Quality of life assessment in paediatric otolaryngology

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Summary

Otolaryngologists are major providers of health care for children. The conditions treated by otolaryngologists can often have wide-ranging effects on a child's health-related quality of life (HRQOL). This thesis contains a review of the available instruments for assessing HRQOL in children, with studies of their applicability in the context of UK paediatric otolaryngology. In addition, two new instruments are described for assessing benefit after an intervention and for assessing quality of family life.

The parents of 274 children referred to otolaryngology clinics with recurrent sore throats, recurrent acute otitis media or otitis media with effusion were asked, depending on the child's age, to complete at least two of the following instruments: the Health Utilities Index, the Child Health Questionnaire, TACQOL and TAPQOL. The responses showed that all the instruments measured HRQOL free from any obvious effect of age, sex or socio-economic deprivation. HRQOL varied predictably with measures of disease severity (such as frequency of sore throats), although the CHQ and TAPQOL lacked sensitivity to the impairments present in otitis media with effusion. Ceiling effects were apparent in many domains in all instruments.

The Quality of Family Life (QOFL) instrument was designed to assess the impact of a health condition on the family. It was applied in the same patient sample described above. QOFL scores were not affected by age, sex or socioeconomic deprivation. Internal consistency was high. More severe disease was associated with greater family impact.

The Glasgow Children's Benefit Inventory (GCBI) was designed as a generic HRQOL measure specifically worded to assess benefit after an intervention and suitable for retrospective application. After initial piloting, it was posted out to the parents of 1777 children who had previously undergone tonsillectomy or ventilation tube insertion. 38% of questionnaires were returned. Correlation between GCBI scores and both technical success of surgery and parental satisfaction were strong. Internal consistency was high and the instrument had a coherent factor structure.

As a result of the work described here, otolaryngologists have information to guide them in their choice of instrument from the wide range available, each suitable for a particular clinical situation.

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Abbreviations

AOM	Acute otitis media
CHQ	Child Health Questionnaire
GBI	Glasgow Benefit Inventory
GCBI	Glasgow Children's Benefit Inventory
HRQOL	Health-related quality of life
HUI	Health Utilities Index
MRC	Medical Research Council
OME	Otitis media with effusion
QALY	Quality-adjusted life-year
QOFL	MRC Quality of Family Life Questionnaire
QOL	Quality of life
RHD-4	MRC Reported Hearing Disability questionnaire
SPSS	Statistical Package for the Social Sciences
TACQOL	TNO-AZL Children's Quality of Life questionnaire
TAPQOL	TNO-AZL Pre-school Quality of Life questionnaire
TNO-AZL	Netherlands Organisation for Applied Scientific Research Academic Medical Centre
TARGET	Trial of Alternative Regimes of Glue Ear Treatment
WHO	World Health Organisation

1 Quality of life assessment

1.1 Why are we interested in quality of life?

Traditionally, outcomes in health care have been expressed in terms of technical measures such as mortality rates, complication rates and laboratory tests. These may not be adequate to describe the patient's own experience of the disease and its treatment. In addition, patients are being encouraged to participate more in decisions relating to their care. The world is changing and greater importance is now being attached to patient-centred outcome measures. The concept of quality of life (QOL) measurement has therefore evolved.

We have moved along the World Health Organisation's hierarchy of impairment-disability-handicap [1] (more recently, but less succinctly, expressed as impairment - activity limitation - participation restriction) from simple assessment of the impairments caused by disease to more sophisticated assessment of handicap in day to day life. For example, recurrent sore throats in a child may impact upon appetite, weight, sleep, behaviour and social interactions, none of which will be directly assessed by simply measuring of the frequency of sore throats. Currently, QOL measurement is used largely in research and economic analyses, but ultimately it may be used to aid decision making for the individual patient [2].

QOL is by definition subjective, however, so its measurement poses a number of problems. This is particularly true in children, who form the subject of this thesis. Any assessment of QOL in children must immediately decide whether to address the child's own response, the parents' response on behalf of the child, or the impact of the child's condition on the rest of the family. Family impact, such as sleepless nights, frequent visits to the doctor and time off work, should not be underestimated in its importance as it may be the prime motivation for the parents to seek medical intervention.

QOL measurement is particularly relevant for otolaryngology, where most interventions are designed to reduce morbidity rather than mortality - patients just want to feel better. The demand from patients for otolaryngology services is high, but the purchasers of health care have questionned the evidence for the efficacy of many of our routine interventions, and even the importance of some of the common conditions we treat. The issue of QOL assessment in children is particularly important, since over a third of all otolaryngological procedures are in children under 14, making otolaryngology the largest provider of surgical care for children in the UK by a considerable margin [3].

1.2 What is quality of life?

"Unlike beauty, which rests in the eye of the beholder, quality of life is inherently an attribute of the...beholdee" [4]. The measurement of something so subjective presents numerous challenges.

QOL is a nebulous concept which is intuitively understood but difficult to define. Its multi-dimensional nature is well articulated in the World Health Organisation's definition of health as "a state of complete physical, mental and social well-being, and not merely the absence of diseases or infirmity" [5].

Overall or global QOL consists of the summary effects of a variety of domains, such as physical, functional, psychological, social and economic [6]. In the context of health care, attention is usually focussed on the areas most affected by disease and its treatment, termed Health Related QOL (HRQOL). The domains of relevance to HRQOL are described in many different ways. For example, Patrick and Erickson [6] describe HRQOL in terms of life expectancy, opportunities for health, perceptions of health, functional status (physical, psychological and social) and impairments. Others include separate domains for role performance (ability to work, do housework, etc) [7] and "resilience and risk" [8]. For the purpose of simplicity, and to keep in line with the WHO definition of health, it is useful to group issues into the three general areas of mental, physical and social, each of which is assessed in terms of both functional status and subjective well-being [9]. This is the most commonly-used and widely-applicable framework for the description of HRQOL.

Some have criticised this approach on the basis that, while functional status can be considered objective to a degree, the subjective sensation of well-being is very dependent on the individual's judgement of how well they are satisfied

with their lot. This judgement will be influenced by their expectations which may be unrealistic. The distinction is drawn between a "capability" approach to measurement based on functional status and a "welfarist" approach which incorporates subjective well-being [10]. However, to disregard subjective well-being completely would be to ignore the relative importance to the individual of their functional impairments [4], and would not be in keeping with the multi-dimensional concept of health described above.

HRQOL is a dynamic concept which varies between individuals in the same health state and in the same individual at different times. The terms HRQOL and health state are often used interchangeably, but here health state is used to refer to a physical assessment of the severity of disease. For example, for a person with a hearing impairment due to the disease otosclerosis, the audiometric pure tone threshold is a measure of health state (disease severity). Two people with the same audiometric results may differ in their HRQOL, as measured by, for example, their ability to function in social situations (functional status in physical and social domains) and any associated distress caused (subjective well-being in those domains).

Health state (disease severity) explains a small part of the variability in HRQOL, which in turn explains only a small part of the variability in overall QOL. There is, therefore, only limited scope for any health care intervention to impact on overall QOL [11].

Quality of life is influenced by many things other than health state (disease severity) and may change over time. Personality (enduring traits which predate the illness) undoubtedly exerts an influence such that, for example, optimistic people cope better with physical symptoms [12]. Expectations change with time, altering the benchmark against which people judge their quality of life. This leads to some patients with treated cancer and chronic diseases reporting better than average HRQOL, as they are now living life to the full and savouring every moment [9].

The way a person reacts emotionally, copes and functions in a given health state is their HRQOL [9, 11]. Indeed, after disease severity, coping style is the most important determinant of HRQOL in children [13].

A relatively new approach is to measure experience and expectations and then define HRQOL as the difference between the two. It has only recently been translated into practical instruments [14-16]. Clearly, a person's HRQOL may be affected as much by something that alters their expectations (counselling, the passage of time, "acceptance") as by an intervention that alters their health state [16].

The dynamic, variable nature of HRQOL is sometimes used to criticise outcomes research as being in some way unscientific. In fact, it is precisely because HRQOL does not vary directly with disease severity that it is necessary to measure it in its own right.

1.3 Why measure HRQOL? Clinical aspects

HRQOL assessment forces the clinician to operate in what may be unfamiliar territory, examining aspects of the patient's day to day life which are not usually discussed. This has the potential to lead to a more patient-centred approach to consultation and decision-making, where the areas of most concern to patients can be highlighted and communication facilitated [2]. However, most existing HRQOL instruments are only useful as research tools, producing aggregate data for a large group of patients in, for example, a clinical trial. We do not currently have instruments which give meaningful, or even interpretable data for any individual patient.

1.4 Why measure HRQOL? Economic analyses

When decisions are to be made about the allocation of limited resources to different aspects of health care, economic analyses can be helpful. The common theme is that the cost of achieving a certain amount of health benefit is compared for two interventions [17]. The key feature of different analyses is the unit for measuring benefit.

In cost-benefit analysis, everything, including survival and HRQOL, is given a monetary value, and an intervention is judged worthwhile if the financial benefits (both health and non-health benefits) exceed the costs. Financial values

for health outcomes may be determined by studies looking at how much money subjects would be willing to pay for an operation, for example.

Where a natural measure is used, such as number of lives saved or cancers detected, one can produce a cost-effectiveness analysis. The conclusions are framed in a way that is easy to understand, such as "£1000 per life saved" or "£300 per cancer detected". Outcomes are not, however, always as clear-cut as life or death. Although it is tempting to use a measure of HRQOL to work out "HRQOL gain per pound spent" the theoretical and statistical basis for this is very weak unless one goes to great length to show that HRQOL is measured on a mathematically interpretable scale. The outcome should also reflect the value that people place on living in a given health state if it is going to be used for economic comparisons [17]. This effectively produces a cost-utility analysis as described below.

In a cost-utility analysis, interventions are compared in terms of cost per "year in full health" achieved. The unit of "a year in full health" is a product of length of life and HRQOL, and the most common unit used is the Quality Adjusted Life Year (QALY). The HRQOL is expressed on a scale from 0 (dead) to 1 (full health) and this is multiplied by life expectancy. The assumption is that 10 years of healthy life are equivalent to 20 years in a state with a HRQOL of 0.5. These values for HRQOL are called health utilities and are defined according to preferences expressed by people from a large population sample. Preferences are measured using time trade-off and standard gamble techniques

to equate a certain length of time in a given hypothetical health state with a longer time in a worse health state [18]. The instruments used to determine the health utilities are called multi-attribute utility scores and differ from other HRQOL measures in that they are weighted using the population preferences and produce a final outcome score on a scale from 0-1. The most commonly used are the Health Utilities Index, the Quality of Well-Being score and the EuroQOL (EQ-5D) [17].

The obvious disadvantage is that the preference-based weightings are derived from how people think they would value life in a given health state, without ever having experienced it. The preferences may not, therefore, reflect the feelings of people actually in that health state. For example, there is evidence that children with physical handicaps give similar ratings of their HRQOL to healthy children [19]. The health utilities are really a measurement of the value that the rest of society places on your life if you have a certain health state. Of course, the health economists would argue that it is society's valuation that matters when society is footing the bill for health care. Equally, most people would justifiably object to being told that their life has been judged to be of little worth to society beause of a health condition.

Other objections that have been occasionally raised to health utility analysis are that the multi-dimensional nature of HRQOL is lost when everything is reduced to a single figure and that utility measures are often insensitive to

small but important differences in health and are, therefore, only usually of use for the study of large populations.

1.5 How do we measure HRQOL?

Essentially, HRQOL is assessed by questionnaires in which patients answer a series of questions about aspects of their day-to-day life.

Many clinicians remain deeply sceptical about HRQOL measurement and its relevance to clinical practice. This is partly due to a feeling that we are trying to quantify something which is nebulous and unmeasurable, and partly due to the unfamiliar nature of the results which have no obvious, intuitive meaning. It is also not helped by the poor quality of many publications purporting to assess HRQOL. In one study of this issue, many publications were criticised for failing to define HRQOL, failing to specify which domains were of interest and why, failing to justify their choice of instrument, failing to distinguish between overall QOL and HRQOL and failing to assess the emotional impact of functional impairments [4]. In addition, many publications which claim to measure HRQOL do so with specifically created questionnaires which have not been subjected to any evaluation of reliability or validity.

Some instruments which purport to measure HRQOL are little more than symptom scores. This is an important distinction to make: symptoms define

health state, but it is the functional limitations and psychological distress that they produce which constitute HRQOL. Instruments which consist solely of a list of questions about symptoms will have spuriously high measures of validity (a demonstration that the instrument measures what it purports to measure – see section 1.7 below). For example, OSA-18 is an 18 item questionnaire which is intended to assess HRQOL in the setting of obstructive sleep apnoea [20]. At least six of these questions are simple descriptions of symptoms, with no assessment of any resulting functional or psychological problem. Since these are the same symptoms which define the presence of obstructive sleep apnoea, the instrument appears to correlate highly with physical findings which are known predisposing factors for the condition. The strongest statistical associations are in the "physical symptoms" and "sleep disturbance" subscales (largely assessments of the presence of symptoms), while the "emotional disturbance" and "daytime function" subscales (more akin to HRQOL as defined above) show poorer association. OSA-18 may be a reasonable measure of health state (disease severity), but it is not measuring HRQOL well.

As another example, Myatt and Myatt have produced an instrument which they describe as a QOL questionnaire to measure pain in children after tonsillectomy [21]. Although produced and evaluated like a QOL instrument, with the usual assessment of validity and reliability (reproducibility and freedom from measurement error – see section 1.7 below), it is not addressing HRQOL at all as defined above, and is really a behaviour-based symptom score. Not every

questionnaire is a HRQOL measure, a point which seems to be lost on some people.

For HRQOL measures to be useful, we must explicitly state what is being measured. Most HRQOL measures cover aspects of physical, psychological and social functioning, but how these areas are addressed and the relative importance attached to them vary widely. There is no consensus or gold standard, but the more comprehensive the range of questions used, the more effective the instrument is likely to be. New measures are being developed in which a "tailor-made" approach is used, the choice of items to be covered being determined by the patient [22].

1.6 Definitions and principles

The following definitions will be used throughout this thesis and represent, I believe, the way that the terms are used most often in the literature on HRQOL.

Generic measures of HRQOL are broad and applicable to a wide range of conditions or treatments, enabling comparisons between the HRQOL produced by different diseases and the benefits of treatments for those diseases. However, any generic instrument will contain few items relating directly to the condition under study. The results will also vary widely due to the effects of items unrelated to the condition of interest. For these reasons, generic

instruments often lack sensitivity to the impairments present in any given condition.

Disease-specific handicap measures, on the other hand, can be made much more sensitive because the scope of questions can be made narrower and more focussed on the condition concerned. It is usual to see these instruments referred to as "disease-specific quality of life measures", which is clearly a contradiction in terms since HRQOL is an overall assessment, effectively the product of all possible disease-related handicaps. Most clinical studies will involve the use of one generic and one disease-specific instrument together.

Direct measures consist of a single global rating, such as a visual analogue scale, to provide an overall estimate for HRQOL. An example from adult practice is the widely used and validated EuroQol visual analogue scale [23]. Such a measurement is obviously easy and quick to do, but is prone to bias because the respondent has to make a judgement based on the combined effects of the various dimensions of QOL. Direct measures are more susceptible to the effects of personality. *Indirect measures*, based on a large number of questions which each address individual areas of HRQOL, produce a more reliable assessment and allow the various dimensions of HRQOL to be addressed separately.

Most instruments are designed so that responses can be added up in some way to produce an overall score. The aggregation of scores from many different domains allows for comparison at the expense of sensitivity, because changes in

individual domains may be masked by changes in other domains. Some argue that summation of scores is illogical when HRQOL is defined as a multi-dimensional concept, and that only domain scores should be reported [24]. No consensus exists as to how much weight should be given to each domain in the overall score, unless a population preference-utility approach is used.

From a practical point of view, the term *item* is used for an individual question, the *scale* is the means provided for answering the question (blank line for free text, Likert rating scale, visual analogue scale, etc), the *domain* is a focussed area of attention made up of a number of items, and the *instrument* is the collection of items forming the questionnaire. If the item responses are added together in some way to produce a single overall value, it is referred to as an *index*, whereas a *profile* preserves the domain structure and cites the results for each domain separately as *sub-scales*.

1.7 Status *versus* benefit measures

HRQOL *status* measurements refer to HRQOL at a particular point in time, whereas *benefit* measures are worded specifically to assess the effectiveness of a particular intervention [25, 26]. Almost always, HRQOL measures are status measures. The clinical situations in which HRQOL assessment is potentially most useful, however, usually involve measurement of change after an intervention.

It is possible to measure change as the difference between two conventional health-related quality of life status instruments, one applied before the intervention, the other afterwards. With this approach, however, it is often difficult to show a change because the small differences produced by the intervention are masked by the large variations in reported quality of life between individuals. In addition, the variance in the post-intervention scores is added to that of the pre-intervention scores when one score is subtracted from the other. Floor and ceiling effects in the response scales may limit the range over which people can report changes in their health.

One other drawback of the before-and-after approach is *response-shift bias* which has recently been demonstrated in children with otitis media in a study using the OM6 instrument [27]. It is apparent from the responses in this study that parents often only realise after surgery that the situation before surgery was worse than they had thought. Pre-operative handicap scores may, therefore, underestimate the degree of impairment and the true benefits of surgery may be hidden.

A measure which is specifically worded with reference to change after an intervention can be much more sensitive to change and free from the effects of response-shift bias. In addition, such a measure can be retrospectively applied to a cohort of subjects who have undergone the intervention in the past, without the need for any questionnaires to be completed before the

intervention. This is particularly useful when attempting to assess the benefits of an intervention which is performed so infrequently as to make prospective data collection impractical.

It is still possible for a direct benefit measure to be affected by expectation bias (a constituent of the placebo response), where parents who have put their child through a (potentially painful) surgical procedure are primed to report some degree of benefit, even where none exists, rather than consciously acknowledge that the procedure was not worthwhile. Studies in adults show that this does not seem to be a major problem in practice, with responses distributed around a value of zero after procedures deemed to have been a technical failure [25].

A post-intervention health-related benefit measure, the Glasgow Benefit Inventory (GBI) [25], exists for use in adults and has been widely adopted for research in various aspects of otolaryngology, including tonsillectomy [28], snoring surgery [29], bone-anchored hearing aids [30], acoustic neuroma surgery [31], rhinoplasty [32] and speech therapy for dysphonia [33].

1.8 Validation of HRQOL measures

1.8.1 General comments

A patient's experience of disease or treatment is inherently subjective and therefore cannot be independently verified. This is often levelled as a criticism of outcomes research, although it should be remembered that many clinical outcomes are simply the subjective opinion of a doctor. Whatever the approach used, it is important to demonstrate that the measure proposed is valid and reliable. However subjective HRQOL may be, its assessment must be as systematic as possible.

A new measure of HRQOL begins with a large number of potentially useful questions, which are reduced in number by excluding any that are ambiguous or difficult to answer, or unable to discriminate between outcomes (i.e. everybody answers the same). Well-established psychometric principles [9] are used to demonstrate both reliability and validity. Reliability is concerned with the precision of measurement and the reduction of random error. Validity is the demonstration that the instrument measures what it is supposed to.

1.8.2 Reliability

Reliability can be assessed in a number of ways. The same test applied a few weeks apart should not give wildly different results unless there has been some

sort of change in the disease (low test-retest variability). Instruments which are administered by trained interviewers should give similar results regardless of who is administering them (high inter-rater agreement). There should also be an appropriate scaling of responses, with no floor or ceiling effects where the wording of items limits the range of possible responses in one or other direction so that very good or very poor health states cannot be reported.

The items that make up the total score should ideally all be measuring aspects of a single coherent concept (HRQOL), so they should be correlated with one another. The degree of correlation (internal consistency) can be assessed with the statistic Cronbach's alpha, which is calculated from the number of items, the variances of the individual items and the variance of their sum [34]. An alpha of zero means no relationship between items, an alpha of one means perfect correlation. For instruments designed to give averaged results for a group (such as in a clinical trial) alpha values of 0.7-0.8 are usually considered adequate, but if the instrument is intended for use in individual patients, some would suggest that higher values (0.90-0.95) are required [34]. If alpha is too high, however, it is likely that many items are redundant.

It does not appear to make a difference to the results obtained when HRQOL instruments are completed at home or in a clinic setting [35].

1.8.3 *Validity*

Validity is dependent on the context in which the instrument will be used, and the clinician should judge a measure primarily on whether it is appropriate to the clinical situation. It is usual to describe validity in terms of content validity and construct validity.

Content validity relates to the appropriateness of the choice of items. Face validity is a common sense assessment of whether questions address the issues concerned and are likely to measure what they are supposed to. It is important to add that questions should be addressing primarily the concerns of patients, and the involvement of patients early in the design of an instrument is invaluable. A comprehensive range of questions will maximise the extent to which the HRQOL measure approximates the real-life experience. The number of questions used, however, must be kept down to a practical level. The questions and their presentation must be suitable for the age range of patients under study, and be suited to their level of reading ability [36].

Construct validity is established by the setting up and testing of hypotheses about how the results of the instrument will correlate with the results of other tests. For example, discriminant validity is shown by the ability to distinguish particular groups of subjects, such as those with mild and severe forms of a disease. Concurrent validity is established when there is significant correlation with other similar measures. Of course, if concurrent validity were perfect, the

new instrument would be redundant unless it were significantly easier to use. Convergent validity is established by showing that the instrument correlates with measures of distinct but related areas, such as disease severity. Divergent validity, on the other hand, is shown if the instrument is shown not to correlate with a measure of something unrelated, such as a behaviour questionnaire or a depression measure.

Responsiveness is the instrument's sensitivity to changes in HRQOL over time. It is essential that an instrument is adequately responsive in any study where the instrument is administered before and after an intervention. Responsiveness may be limited by floor and ceiling effects.

The originators of an HRQOL instrument may choose to define a number of domains each consisting of a subgroup of items that all relate to a particular aspect of the patient's experience. Items within a domain (for example, "psychological effects") should correlate statistically with each other more than with items in another, unrelated domain (such as "mobility") or with the instrument as a whole. Alternatively, relationships between all the items can be studied with a statistical technique known as factor analysis which will determine which groups of items cluster together statistically. These groups of items are called factors.

Because validity is context-specific, cultural differences exist which affect the responses given, making it difficult to compare responses from different

countries or ethnic groups [37]. The factor structure of an instrument may vary when it is applied in different countries [38, 39]. Indeed, the whole concept of "quality of life" may differ. For example, in Germany, it has been suggested that the term "quality of life" carries negative connotations by its association with "value of life" and euthanasia during the Nazi regime [39]. When an instrument is to be used in another country, simple translation is not enough: the instrument must demonstrate its validity in the new setting. Back translation, committee review, piloting and re-examination of score weighting are recommended [40].

Validity is not a fixed property of a measure, but rather something dependent on the specific purpose or setting. It is, therefore, meaningless to refer to a "validated instrument". It would be more appropriate to say that the instrument appears to be valid in a particular setting. This does not, of course, mean that it will be valid in any other setting.

2 HRQOL in children

2.1 Practical issues

Medical advances mean that mortality, the traditional outcome measure for medical intervention, is only encounterd with any regularity in certain areas of paediatric medicine such as very low birth weight neonates, oncology and transplantation [8]. Even in these areas, the preservation of life often comes at the cost of substantial lifelong morbidity. For most of paediatric healthcare, even more than for adults, the day-to-day burden of morbidity is what matters. The assessment of HRQOL in children is, therefore, relatively more important. It is also much more difficult.

In any study involving HRQOL assessment in children we must be clear from the outset about whether to measure the child's own response, the parents' response on behalf of the child or the impact of the child's condition on the family. Studies suggest that parents and children produce different responses, but that both assessments have a place and should probably be viewed as complementary [41, 42]. Studies of the correlation between child self-rated HRQOL, parent rated HRQOL, physical symptoms and objective measures show that parental proxy responses add little extra information when dealing with children over 11 years, but are a useful adjunct for younger children [43].

Proxy reports of functional status, such as ability to walk or dress, are to a large extent "objective", whereas proxy reports of emotional distress in a child are more prone to reflect the values of the parent. Children may have more knowledge of their recent functional status than their parents, and more interest in physical aspects of their illness [44]. These effects can be seen in some studies, where parents tend to place greater emphasis on the emotional effects on the child, whereas children are more concerned with the physical effects [45, 46]. Studies on proxy reports in adult health also show that they are more likely to agree with the patient's own assessment for "hard" (physical) data than for subjective (emotional) evaluations [47-51], and the same seems to be true for parents and children [42, 52]. However, even the accuracy of parental estimates of "hard" data, such as a child's hearing level, can be poor [53]. On the other hand, parent reports are more reproducible and responsive to change, and thus may be more suitable for longitudinal studies where measures are repeated [54].

The use of adult instruments in a paediatric setting is unfortunately common, and not recommended for a number of reasons [51]. It is clear that HRQOL instruments designed for adults are unsuitable for children because they may not address the appropriate areas of concern, and do not frame responses in the context of the child's age and developmental stage [51, 55]. They may also be too long, or too complicated for children to read [51]. The domains of interest in children are the same as in adults, if we use the broad definitions previously described – physical, social and psychological, each assessed for functional level

and subjective well-being [9]. However, within these broad domains, the specific areas of interest will be very different to those in adults.

Children develop rapidly, so functional status must be assessed relative to what would be expected for a child of a certain age. This use of a hypothetical "normal" reference point makes it possible to produce an instrument which is applicable to a wide age range [56]. An alternative approach is to produce different HRQOL instruments for children of different ages, although this will limit their use for longitudinal studies where children are followed up for a long period of time.

Older children and adolescents may spend more time with their peers than with the family, and peer relationships may provide a closer analogue for many adult QOL domains than the family setting [57]. For younger children, the family remains central to their day to day experience and development.

Most instruments for use in children specify a lower age limit, based on the child's ability to read the questions, understand the concepts and make an appropriate judgement. To a certain extent, limited reading skills can be compensated for by using simple language and a "smiley faces" rating scale [58], but the child still needs to be able to read. Limitations in abstract reasoning and a tendency to concentrate on recent events can also affect a child's ability to report HRQOL accurately. Until recently, no self-report measure existed for children below 8 years of age. Any such measure would have to be quite

innovative in its design, not least because young children may not be able to read well enough to complete a printed questionnaire, and their short attention span limits the number of questions which can be asked. Trained interviewers are an expensive and labour intensive answer to the problem, but a better answer may be to use an animated computer programme to gather the child's responses. This approach has been used successfully for children as young as 5 years to develop a disease specific HRQOL measure for inflammatory bowel disease [59] and a recently-reported generic HRQOL measure for children as young as 6 [15]. Although promising, data on the latter ("Exqol") are limited to a single preliminary report and further results are awaited.

2.2 Impact on the family

Living with a person who has an illness has an impact on the rest of the family. This may be shown in the need to spend time with the affected person, including time away from work. It may disrupt family life by causing sleepless nights, changes to routine and limitations on activities such as holidays and leisure pursuits. It may also cause worry, stress and financial expense. The impact of a disease on the rest of the family is very real, and therefore something that should be amenable to some sort of measurement.

This begs the question, of course, of why we should need to measure it. Decisions in health care are, in certain situations at least, determined by the

family as much as by the individual. This is certainly true for vulnerable groups such as the elderly, the learning-impaired, those with physical disabilities and children. For these groups, the decision to seek health care intervention, and the nature of the intervention chosen, may reflect the impact the disease has on the family more than the impact of the disease on the individual. For example, many hearing-impaired elderly patients are persuaded to attend for provision of a hearing aid because their family members are frustrated at the difficulty they have in communicating, even though the elderly patients themselves may not perceive much of a problem [60].

In paediatric medicine in particular, the concerns of parents are the prime motivating factor in the decision to seek health care intervention, especially for young children. The decision to undertake tonsillectomy in a child with recurrent sore throats, for example, will be influenced by the parents' concerns, sleepless nights and need to take time off work as well as by the perceived effects of the sore throats on the child. The extent to which such decisions are influenced by the child's perceived quality of life as opposed to the impact on the family is currently unknown, but there is evidence that parental factors play a large part in the decision to seek medical attention [61]. Teasing out such influences will require instruments to measure child and family impact separately.

Such research is of particular importance in paediatric otolaryngology, where there are many interventions which have high levels of public demand, but for

which the evidence of efficacy is perceived to be poor among health care purchasers. Such procedures as tonsillectomy and ventilation tube insertion, the two most common surgical procedures performed in children, can be described in this way. Parents may hold differing beliefs from health care professionals regarding the diagnosis, prognosis and treatment options for common ear, nose and throat conditions [61]. The number of tonsillectomy and ventilation tube operations varies widely from one place to another in the UK without obvious reason [62, 63]. Research to understand the variability in decision making regarding these operations is essential to move towards a more equitable and efficient service, and research into the effect of family impact on decision making will be key.

2.3 Existing generic HRQOL measures

2.3.1 General comments

Methods for the measurement of HRQOL in adults have been developed over many years. Instruments such as the Medical Outcomes Study Short Form 36 (SF 36) and the Nottingham Health Profile have been extensively validated and are widely known and used. HRQOL measurement in children is a more recent area of development, and no instrument can be seen as a "gold standard" yet.

Attempts have been made to produce generic HRQOL measures for children, to provide a benchmark for comparison between different diseases. The need to keep things simple and concise for use in children conflicts directly with the need to cover a broad range of issues that may impact on HRQOL. Some measures which are described as generic are, in fact, more suitable for use in certain groups of children (e.g. cancer) and may not be sensitive to the HRQOL issues present in children with other complaints. This is an issue of face validity.

The ideal instrument would be child-centred, easy to use and suitable for completion without the need for a trained interviewer, as well as being sensitive to small differences in HRQOL and applicable to a wide range of disease states and ages.

2.3.2 *The Child Health Questionnaire*

The Child Health Questionnaire (CHQ) is a generic measure of HRQOL which has been specifically designed for use in children [56]. A UK version, with minor modifications to the American spelling and wording, has been produced and tested [64]. The parent form is scored by parents on behalf of the child and is divided into 15 domains, namely Global Health, Physical Functioning, Role/Social Limitations Emotional, Role/Social Limitations Physical, Bodily Pain, Behaviour, General Behaviour, Self Esteem, Mental Health, General Health Perceptions, Parental Impact Time, Parental Impact Emotional, Family

Limitations in Activities, Family Cohesion, Change in Health. These are summarised into a Physical summary score and a Psychosocial summary score.

The original version has 98 questions, but a shorter version with 50 questions (CHQ-PF50) is the most widely used, and a 28 question version (CHQ-PF28) is also available. The CHQ has been used in various contexts including asthma, epilepsy, chronic renal failure, oncology, arthritis, attention-deficit-hyperactivity disorder and cystic fibrosis (information from http://www.qlmed.org/CHQ/). It is designed for use in children aged 5 years or older [56]. A self-report version for children aged 10 or more (CHQ-CF87) has also been developed in parallel. It has 87 questions with the same domain structure as the parent forms.

Responses are given on a variety of categorical rating scales, with between 4 and 6 categories. The responses with an "excellent" to "poor" response range are recalibrated to provide a better approximation of an equal interval scale. Each response is then multiplied by a weighting factor before they are all summated. The actual calculations are lengthy and complex and are only practical if done by computer.

The CHQ-PF50 has been validated on a random sample of 391 US children aged 5-18, stratified for age, sex and parental employment. Samples of children with attention deficit-hyperactivity disorder (83 children), asthma (3209 children), epilepsy (34 children), psychiatric disorder (82 children) and juvenile chronic

arthritis (74 children) have also been used to validate the instruments. Differences in scores were found according to the age, sex and ethnicity of the child, and the socio-economic status of the parents. For the CHQ-PF50, tests of internal consistency showed Cronbach's alpha values of at least 0.7 for all sub-scales except General Health (0.66) in the population sample, and at least 0.56 for all sub-scales in the children in the clinical groups. The summary measures, Physical and Psychosocial, had alpha values of 0.93 in the population sample, and ranged from 0.84 to 0.97 in the clinical samples. For the CHQ-PF28, the results were not so consistent, with alpha values of 0.89 for the population sample. Factor analysis confirmed the validity of using the two summary scores. Construct validity was confirmed by comparing the scores between the population sample and the clinical groups [56].

The CHQ-PF50 has also been used in a general UK population sample by workers at the MRC Institute for Hearing Research in Nottingham [38]. They found poorer internal consistencies for three subscales compared to the reported USA data (Parental Impact Emotion, Mental Health and Global Health Perceptions), but overall they found the CHQ was likely to be suitable for use in a UK population.

2.3.3 *The Health Utilities Index*

The Health Utilities Index (HUI) is a generic health utility measure which was originally developed in Canada in the 1980s [18]. The original version, referred to

as mark 1, was designed to assess outcomes in very low birth weight neonates. The system comprised four domains (physical function, role function, social-emotional function, health problems). Subsequently, 84 parent and child pairs evaluated a list of possible attributes (decided on the basis of a literature search) to determine which were most important to them, using a visual analogue scale and time trade-off techniques. These preferences formed the basis for weighting the items in the HRQOL assessment to produce utility scores.

The HUI mark II system was designed for use in the survivors of childhood cancer, and a seventh domain relating to future fertility was added because of this [18]. Preferences were elicited from 293 parents using a visual analogue scale and the standard gamble, and these were used to weight the items. Some problems were identified, particularly with a lack of independence between certain items, and a revised version (mark III) was produced.

The mark III was designed to be suitable for use in general population health surveys. The most problematic area in the mark II was related to self care, and in the mark III this was replaced with an item relating to dexterity. Sensation was broken down into speech, hearing and vision, and the fertility item was omitted. Weightings were produced from 504 adults using a visual analogue scale and the standard gamble. The resulting eight-domain HUI mark III has been used in a number of large population studies in Canada, from which norms have been calculated [18]. Forms are available for self-report, telephone interview and face-to-face interview, and for parent/proxy and child reports.

Although not specific to children, the HUI mark III has potential to be particularly useful in paediatric otolaryngology because it is one of the few instruments to address hearing and speech specifically (in the mark II they are assessed together with vision as "sensation"). The attribute levels described for the HUI mark II are referenced to what would be expected for a normal child of the same age, whereas those for the HUI mark III are not. This is particularly important for vision ("can read newsprint"), speech ("can be understood by strangers"), mobility ("can walk without assistance") and self-care ("can eat, dress, bathe and use the toilet without assistance"), where children under 4 would not usually be able to perform these functions unaided. The originators suggest a lower age limit of 6 years on the use of the HUI mark III. The mark II should not have a lower age limit.

The HUI mark III comprises 15 questions, mostly about aspects of physical functioning. There is only one item relating to subjective well-being ("being happy") and social issues are not addressed at all. For each item, the respondent is asked to choose from a list of 4 to 6 choices the one that best describes their current ability to function. For example, the respondent may answer a question about "your child's ability to see well enough to read ordinary newsprint" by selecting the description "able to see well enough with glasses or contact lenses". From these descriptions, it is possible to calculate health utility scores, which reflect the value placed on life in such a state by a large group of normal people in the population. These utilities are calculated

for each of 8 separate areas, known as single-attribute utilities (vision, hearing, speech, ambulation, dexterity, emotion, cognition, pain) and for life overall, known as a multi-attribute utility. Each is expressed as a number between 0 and 1, with 0 representing death and 1 perfect health. The HUI questionnaire also includes a 5-point Likert-type rating scale of overall health, but this is not used for calculating utilities.

The HUI marks II and III have been used in a number of clinical studies, and have been found to be able to discriminate between groups with a different burden of illness. There are limitations, however, in relation to the narrow definition of HRQOL used (primarily relating to physical impairment) and the scope of the items [65]. While it is clearly advantageous to have an instrument that is short, and therefore easy to complete, having only 15 questions can also be seen as a drawback and has been blamed for the poor discriminative ability shown for the HUI marks II and III in children with asthma [66].

Reliability of the HUI marks II and III has been demonstrated, with a high degree of test-retest agreement [18, 65, 66]. A degree of discriminant validity for the mark II has been shown between very low birth weight and normal children [19], and children with cancer on and off treatment [65].

A study from Canada has compared the CHQ-PF50 with the HUI marks II and III [67]. 244 survivors of childhood cancer completed both instruments. Correlations were moderate or strong for the following sub-scales: CHQ bodily

pain and HUI mark II/III pain (Spearman correlation coefficient 0.58 for HUI mark II and 0.60 for HUI mark III); CHQ physical functioning and HUI mark II mobility (0.45) and HUI mark III ambulation (0.45); CHQ mental health and HUI mark II/III emotion (0.64 and 0.54); CHQ general health scale and HUI mark II/III global utility (0.43 and 0.44); CHQ general health single item and HUI mark II/III global utility (0.38 and 0.42). This high degree of convergent validity is interesting given the completely different underlying philosophies (mutli-dimensional, including psychosocial versus strictly functional) and scoring methods (summation of Likert scales versus population preference-based utility), but it has subsequently been confirmed in a study from the Netherlands comparing CHQ-PF50 and HUI mark II in 467 schoolchildren [68].

2.3.4 *TACQOL and TAPQOL*

The TNO-AZL (Netherlands Organisation for Applied Scientific Research Academic Medical Centre) Questionnaire for Children's Health-Related Quality of Life (TACQOL) exists as a parent form (TACQOL-PF) and a child form (TACQOL-CF), both suitable for children aged 6-15 years [24, 69]. A version for the parents of pre-school children (TAPQOL) has been developed [70], as has an adult HRQOL instrument (TAAQOL).

For the parent and child versions of TACQOL, questions were formulated on the basis of expert discussions and a literature search, then modified after a pilot study of 77 parents [69]. The questions in the child and parent versions are

essentially the same, the only differences being in the wording used. There are 63 questions in total for each. Questions were grouped in seven sub-scales, Body, Motor, Cognition, Autonomy, Social, Positive Emotions and Negative Emotions. The modified questionnaire was then used to collect population data from 1789 parents, and 1159 children aged 8-11 [71]. The results confirmed the validity of the structure and scoring of the questionnaire, and showed differences between the scores for healthy children, children receiving medical treatment and the chronically ill, although the effect sizes were small.

The great strength of the questionnaire is that functional status and its emotional impact are measured separately, and the authors justify this distinction by showing that 43% of reported functional limitations were not associated with negative emotional reactions. Scores for each question are based on the functional limitation weighted for its emotional impact. The Positive Emotions and Negative Emotions sub-scales are not weighted any further. Scores for each sub-scale are calculated by simple addition of question scores, giving scores from 0-32 for the 5 weighted sub-scales, and scores of 0-16 for the Positive Emotions and Negative Emotions sub-scales. No overall score is calculated.

Comparison between the child self-reports in TACQOL-CF and the parent reports in TACQOL-PF show that significant differences were present [41]. Children reported poorer HRQOL than their parents for the Body, Motor, Autonomy, Cognitive and Positive Emotions sub-scales. Age, gender and

HRQOL score all had small, variable effects on the degree to which parent and child reports agreed. Both reports appear to be valid in their own way, and should be seen as complementary. It is noticeable, however, that agreement is poorer for those attributes which are less easily observed, such as mood, pain and social functioning [52].

TAPQOL was designed along the same lines as TACQOL, and is presented in the same format. 43 items are included in 12 domains, based on expert opinion, literature review and discussions with parents. The questionnaire was tested on 121 parents of preterm children and 362 parents of normal children from the general population [70]. Cronbach's alpha for most scales exceeded 0.6. The preterm children, and children from the population sample with chronic diseases scored lower than the healthy children from the general population.

TAPQOL was used in a study of HRQOL in children born preterm: there were significantly poorer scores in those born at 32 weeks gestation or less compared with a reference sample of children born at term [72].

2.3.5 *Other instruments*

PedsQL was developed in California and has 23 questions in four Generic Core Scales (Physical, Emotional, Social, School) and is designed to be administered alongside a number of PedsQL Disease-Specific Modules to produce a comprehensive but sensitive HRQOL measure. Although the initial modules were cancer-orientated [73], it is expected that a range of disease-specific modules will be made available. It can be completed either by the child (aged 8 or above) or by the parent, and internal consistency is high for both child- and parent-report (alpha 0.88 and 0.9 respectively). The scores correlate with markers of disease burden and distinguish healthy from ill children [74]. It shows great promise, but use is as yet limited and otolaryngology-specific modules are not yet available.

KINDL is a German generic measure of HRQOL, which has been piloted in a small number of children [75]. It comprises 40 questions in 4 domains (mental, physical, social, everyday life) and has been used in children aged 9-11 years. An English translation is available, but it has yet to be properly validated. A comparison between KINDL and TACQOL showed that the correlation between the two was low, even for scales intended to measure comparable concepts [71]. This may in part be due to a different time frame for KINDL (previous 1 week) compared with that for TACQOL (previous 4 weeks). KINDL has a high degree of correlation between its subscales, much more so than TACQOL, which has been taken to suggest that KINDL is only measuring

a single aspect of HRQOL ("general health"), while TACQOL fits better with a multidimensional concept of HRQOL [71].

The Generic Children's Quality of Life Measure (GCQ) has been developed recently in Nottingham. It is a self-report measure, and has been used by children aged 6-14 years [14]. It takes a novel approach, asking children to rate on a five-point pictorial Likert scale how they see themselves, and how they would like to be, in relation to 25 items. HRQOL is defined here as the difference between how children see themselves and how they would like to be [16]. The items were chosen based on discussions with children about the issues they consider important in determining HRQOL, and cover physical, psychological and social domains. In a sample of 720 normal school children, the GCQ produced a wide range of scores with a normal distribution. The reliability coefficient was high (Cronbach's alpha 0.74-0.78), and there was no significant effect of age, sex or socioeconomic class [14].

16D and 17D have been developed for use by children in certain age groups, namely 8-11 years for 17D, and 12-15 years for 16D [76, 77]. They cover psychological, social and physical issues, but most areas covered are of little relevance to otolaryngology. Use so far has been limited to the original descriptive studies. 17D has some nice pictures accompanying the questions. The comparability of 16D and 17D is unclear, limiting their use to either of the specific age groups mentioned, which may be a problem for longitudinal studies

The Quality of Well Being (QWB) scale was designed as a health utility measure for chronically ill adults. It has had some limited use in children with cancer [78]. It is cumbersome and needs to be administered by a trained interviewer. It lacks sensitivity to disease status, especially at low levels of impairment, and is generally felt unsuitable for paediatric use [79].

The RAND Health Insurance study was the first attempt to systematically address the issue of HRQOL in children in the context of a population study of the prevalence of disability [80]. It is not sensitive to changes over time, to the HRQOL impairments of children with chronic disease without significant physical handicap [81], or to different levels of dysfunction within a population [44].

CHIP-AE is a self-administered instrument for use in adolescents [82]. It is not designed for use in children below 11 years of age.

Functional Status II Revised (FS-IIR) exists in a 43-item full version and a 14-item short form [81]. It is administered by a trained interviewer, which limits its usefulness. The instrument caters for a wide age range (birth-16 years) by providing panels of questions for each age group.

Wiklund and co-workers [83] have produced a self-administered 34 item generic instrument which specifically measures psychological well-being in children aged 9-13, with no attempt to address functional status or physical symptoms.

It was originally designed for use in children with short stature, but may be useful in other situations where physical symptoms are minimal.

2.4 Comment

The assessment of HRQOL poses particular practical problems in children and this field of study has been slower to develop than in adult medicine. Nonetheless, a variety of instruments now exists with a wide range of formats and very different theoretical bases. A small number of these child-specific instruments have accumulated enough use in different clinical settings to demonstrate their potential as useful generic HRQOL assessment tools. Their relevence to otolaryngology, however, remains to be demonstrated.

3 HRQOL assessment in paediatric otolaryngology

3.1 Practical issues

Children under 14 years make up over a third of all patients seen by otolaryngologists in the UK [3]. These children differ in many ways from the children seen in paediatric medical clinics, and the choice of HRQOL measures to be used should take into account their particular characteristics. Instruments designed for chronic, fairly stable conditions (for example, the survivors of childhood cancer) may not be appropriate for use in conditions which include relapsing episodes of acute illness, interspersed with long periods of being well (for example, recurrent acute sore throat).

The majority of children seen by otolaryngologists suffer from non-life-threatening chronic conditions such as recurrent sore throats, recurrent acute otitis media, chronic rhinosinusitis and otitis media with effusion. In specialist centres, children with congenital anomalies of the head and neck, airway disorders and congenital or perinatal-acquired hearing impairment will also be seen. These conditions tend to be most common in the pre-school age group. Many of these children will have communication difficulties either because of hearing impairment, or anatomical problems affecting speech. Young age and communication difficulties make child self-reported HRQOL difficult.

Generic HRQOL instruments vary in their face validity for use in otolaryngology. The HUI, for example, has questions which specifically address hearing, and it is able to discriminate well between adult patients with varying degrees of hearing impairment (Q Summerfield, MRC Institute for Hearing Research, personal communication). It does not, however, have specific questions which address the problems associated with, for example, recurrent sore throats, so it is not clear that it will be sensitive to the HRQOL impairments of all common otolaryngological disorders.

3.2 Generic HRQOL measures

There is little experience in the use of generic instruments in this specialty.

CHQ-PF28 was used in a series of 21 children undergoing sinus surgery for chronic rhinosinusitis. They showed significantly poorer Physical summary scores (but not Psychosocial summary scores) compared with published norms. Scores in some domains were even worse than those reported in juvenile chronic arthritis, asthma, epilepsy, psychiatric disorders and attention-deficit hyperativity disorder [84].

A small study of the HRQOL impact of congenital craniofacial anomalies used CHQ-PF28: the 27 children studied with cleft lip and/or palate reported Physical and Psychosocial summary scores within the range of normal, as one

would expect after a successful repair. Those with more serious anomalies (for example, syndromic craniosynostosis such as Crouzon's and Apert's) reported significant impairments [85].

CHQ-PF28 has been used to study 55 children with recurrent sore throats and/or obstructive sleep apnoea, showing significant impairments in all domains except Mental Health and Self-Esteem [86]. The small number of children studied, and their widely differing clinical problems, make this study of limited value. However, the CHQ-PF50 has more recently been used in a study of 298 children undergoing home polysomnography in the assessment of obstructive sleep disorders. Significant impairments in both Physical and Psychosocial summary scores were identified. The degree of HRQOL impairment correlated with disease severity on polysomnography, but impairments were present even for those with mild obstructive sleep disorders [87]. The CHQ-PF28 was used before and after adenotonsillectomy for sleep disordered breathing in a study of 55 children. Physical summary scores were improved after surgery, but Psychosocial summary scores were not [88].

The HUI has been used in the USA for cost-utility analysis of cochlear implantation in profoundly deaf children. A cost of $5,197 per QALY was calculated, which translates to a total saving of £53,198 per child when all costs (including educational expenses) are considered [89].

A Dutch trial of ventilation tube insertion after early screening for otitis media with effusion in infants used the TACQOL questionnaire to assess HRQOL benefit from surgery [90]. No benefit was shown, and this was taken to mean that surgery in this group does not improve HRQOL. However, it could also mean that TACQOL is not sufficiently sensitive to the impairments present in young children with otitis media with effusion, or to the changes in HRQOL produced by surgery. The choice of TACQOL as the outcome measure may not have been ideal for these pre-school children, as the questionnaire is designed for use in children over 6 years of age (presumably, TAPQOL was not available at the time of the trial).

A generic benefit measure has yet to be developed for use in children. The Glasgow Benefit Inventory, although designed for use in adults, has been used in a group of children (mean age 10 years) who had undergone tonsillectomy, and the resulting data were used as part of the instrument's validation [25]. The instrument was completed by children in 5 cases, and by parents in 47. It was able to distinguish between those who benefitted from surgery but who still suffered some sore throats, and those who were cured.

3.3 Condition-specific handicap measures

Few condition-specific measures are available in paediatric otolaryngology, and of these, only OM6 has been widely used and tested beyond its original description.

3.3.1 Rhinitis

The Paediatric Rhinitis Quality of Life Questionnaire [91] was developed using questions based on a literature review and discussions with 34 Canadian children with seasonal allergic rhinitis. The draft questionnaire was then piloted on 75 children (mean age 9.8 years) in Texas also suffering from allergic rhinoconjunctivitis. The questionnaire has 23 questions in five domains (nose symptoms, eye symptoms, practical problems, other symptoms and activity limitations). Responses are given on a seven-point rating scale, with a 7 day reference frame. The questionnaire is administered by a trained interviewer. The results show good internal consistency and convergent validity with a symptom diary. The instrument also discriminates those who have responded to treatment from those who have not.

A 5-item questionnaire has recently been described with a format very similar to that of OM6 (*vide infra*). Each item covers a range of symptoms, and is scored positively on a scale of 1-7 if any of the symptoms is present. The symptom-cluster items are sinus infection, nasal obstruction, allergy symptoms, emotional

distress and activity limitations. Initial data suggest good responsiveness to change after treatment [92].

3.3.2 *Otitis media*

OM6 is a beautifully short instrument whose great advantage is speed and ease of use. It has been developed for use in children with recurrent acute otitis media and otitis media with effusion [93]. Six domains (physical suffering, hearing loss, speech impairment, emotional distress, activity limitation, caregiver concerns) are each represented by a single question. The question gets a positive response if any of a list of symptoms is present. Answers are given on a seven-point categorical scale, and a total score is calculated by taking the mean of the six domain scores. A visual analogue scale is also included for a global assessment of ear-related QOL. In 186 children with ear symptoms, test-retest reliability was high, and the correlation between the overall score and the global assessment on the visual analogue scale was high [93]. In a prospective study of 248 children undergoing ventilation tube insertion for otitis media with effusion, OM6 scores before and after surgery showed a significant increase, demonstrating the instrument's sensitivity to change [94]. Similar benefits from ventilation tube insertion were shown in a pilot study of 14 children in Liverpool [95] and in a study of 72 children from the Netherlands [96]. Children with 4 or more episodes of otitis media per year score significantly worse than those with only 2 or 3 episodes per year [97]. The grouping of symptoms into only six questions, however, is likely to introduce a risk of bias

and a lack of sensitivity. In addition, it could be argued that many of the items are simply symptom descriptions without any attempt to assess the impact of the symptom on the child, and that OM6 is, therefore, more of a symptom score than a handicap measure.

A group from Florida has expanded the items in OM6 into a series of 22 separate questions and renamed the result the Otitis Media Outcomes-22 (OMO-22). The format was otherwise unchanged. In a series of 123 children undergoing ventilation tube insertion OMO-22 was found to have low test-retest variability and good internal consistency (alpha 0.85). The instrument was able to distinguish healthy children from those with otitis media and was responsive to change after ventilation tube insertion [98].

Alsarraf and co-workers have developed three parallel instruments for use in children with recurrent acute otitis media [99]. The Otitis Media Clinical Severity Index (OM-CSI) is a 10-item instrument detailing symptoms and signs, for completion by the treating doctor. The Otitis Media Functional Status Questionnaire (OM-FSQ) is completed by parents and is a 14-item disease-specific HRQOL instrument based on the Functional Status II-R (*vide supra*). The Otitis Media Diary (OM-D) is also completed by the parents, who are asked to record the presence and severity of ear symptoms, time spent caring for the child and medication required. The instrument battery was piloted on 25 children with otitis media and 26 healthy controls. Internal consistency and test-retest reliability for the scores are high. Convergent validity has been

shown with physician ratings of severity and parental play ratings. Large change scores after treatment have also been reported, but numbers are small and further experience with use is required.

The Trial of Alternative Regimes for Glue Ear Treatment (TARGET) was a large, multi-centre randomised controlled trial conducted in the UK by the Medical Research Council (MRC) and which was specifically designed to have broad outcome measures. At the time of its inception, no well-validated generic instruments for assessing HRQOL in children were available, and the disease-specific measures all had significant drawbacks. A new outcome measure was therefore developed and validated in parallel with the trial. It was designed to be specific to otitis media with effusion, parent-reported and suitable for children aged 3-9. The instrument includes indirect (item-based) and direct measures. The direct measures comprise three visual analogue scales, one each for the child, the parent and the family. The indirect assessment includes 5 items on general health, 62 items on behaviour and 16 items on parental QOL. Test-retest reliability, internal consistency and responsiveness to change were high. Discrimination between clinical and normal reference samples was good, and convergent validity with appropriate CHQ sub scales was high [100]. Ventilation tube insertion was shown to produce a significant improvement in HRQOL over the first year, with no sustained effect at two years. The magnitude of the effect of ventilation tubes was equivalent to 0.4 of the standard deviation of the overall distribution of the scores [101].

3.3.3 *Sore throats and sleep apnoea*

Originally named OSA-20, an instrument was developed to assess the HRQOL effects of obstructive sleep apnoea [20]. Twenty questions in 5 domains (sleep disturbance, physical symptoms, emotional distress, daytime function and caregiver concerns) were selected by the authors on the basis of personal experience, and piloted on the parents of 61 children aged 6 months to 12 years (median 4). All the children had enlarged tonsils and adenoids and a history of loud snoring with disturbed sleep. The internal consistency of the questions was high. Correlation between domain scores and the results of polysomnography and physical examination were weak and variable, especially for the emotional distress and daytime function domains. Two questions were excluded because they were rarely relevant, and the instrument renamed OSA-18. A subsequent study of OSA-18 in 64 children undergoing adenotonsillectomy demonstrated moderate to large changes in domain scores after surgery [102].

OSD6 is a 6-item instrument designed to assess the impact of obstructive sleep disorders on children and their families. It is structured in a very similar way to OM6, and has similar domains (physical suffering, sleep disturbance, speech and swallowing, emotions and stress, activity limitations and caregiver concerns). Its initial study showed low test-retest variability and good internal consistency (alpha 0.8), but associations with estimates of tonsil size and nasal airflow were poor [103]. A subsequent study by the same author has shown OSD6

to be responsive to change after adenotonsillectomy, thereby confirming the significant positive effect of surgery on HRQOL [104].

A disease-specific HRQOL measure for children with adenoid and tonsil disease (nasal obstruction, recurrent acute sore throat and obstructive sleep apnoea) has recently been reported [105]. Items were chosen by an expert panel, and reduced in number by piloting on a group of 34 parents. Fifteen items are grouped in six domains (airway/breathing, infections, eating/swallowing, health care utilisation, cost of care, behaviour). Further pilot work on 158 parents showed high internal reliability for items in each domain, and high test-retest reliability. Convergent validity was shown with CHQ-PF28 and clinical data. This instrument is in its early stages, and further validation data are required.

A non-randomised study on a cohort of children who had undergone surgery for obstructive sleep apnoea (either tracheostomy or aggressive craniofacial surgery) was reported in which the main outcome measure was quality of life. This was assessed with a disease-specific instrument (OSA-QOL) developed specifically for the study [26]. The instrument consisted of 76 items grouped into 3 domains (health and sleep, medical visits and costs, psychosocial). The items were chosen on the basis of discussions with families of children with refractory obstructive sleep apnoea. Answers were given on a five point Likert rating scale. For each item, two answers were requested, one referenced to "before surgery" and one to "after surgery", giving the instrument the characteristics of

a post-intervention benefit questionnaire. Not surprisingly, therefore, it proved to be responsive to change, as well as having acceptable inter-rater agreement. A difference was shown between the tracheostomy and craniofacial surgery groups, but only after cases with poor clinical outcomes were excluded from the craniofacial group. This clearly introduced an immense bias. Only 44 families filled in the questionnaire, and further validation is required.

3.3.4 Larynx and trachea

The Pediatric Tracheostomy Health Status Instrument is a condition-specific measure developed for the assessment of the effect of a tracheostomy on the child and family. Item selection was based on literature review, expert opinion and parent/caregiver focus groups. Its initial study was in 154 self-selected families contacted via an interactive website offering information on paediatric tracheostomy. The initial results suggest good internal consistency (alpha 0.91), good correlation with global ratings of the child's general health and caregiver's quality of life, and the ability to distinguish subpopulations with and without major comorbidities. The instrument was designed with four domains: physical symptoms, medical visits and costs, stress and coping from the child's perspective and stress and coping from the caregiver's perspective, and this structure was confirmed with factor analysis [106].

The same author has developed the Pediatric Voice Outcome Survey by modifying an adult instrument, the Voice Outcome Survey. The modifications

made were to rephrase the items to allow a parent-proxy response. In addition, a single item on swallowing was omitted as it had poor correlation with the rest of the items. The resulting instrument was piloted on 108 self-selected families via the interactive tracheostomy website. The instrument proved to have moderate internal consistency. Children with a current tracheostomy had poorer scores, as one would expect, than those successfully decannulated [107]. A subsequent study by the same author contains reference data from a general paediatric otolaryngology population together with test-retest reliability data [108].

3.4 Quality of family life

The PAR-ENT-QOL instrument has been developed to measure the impact of childhood ear, nose and throat infections on the parents [109]. Although an English translation is available, the questionnaire has only been validated in French, Italian, German, Czech and Portuguese. It consists of 17 questions, plus a global assessment of QOL, all rated on a five-point Likert rating scale. The questions were based on a literature search and discussions with parents. It has been piloted on the parents of over a thousand children with recurrent ear, nose and throat infections. Three questions were subsequently excluded, two because they were relevant to only a minority of those tested, and one because it did not correlate with the global QOL rating. Principle component analysis was used to divide the remaining questions into two sub-scales, Emotional

Score and Daily Disturbance Score. These two sub-scales and the total score all correlated with the number and type of infections, and their socio-economic consequences such as number of days lost from work, eviction from day-care and need for babysitting help.

Milczuk and Johnson reported a study on the effects on the family of caring for a child with stridor due to laryngomalacia [110]. The HRQOL assessment measure was developed *ad hoc* for the study, and no details are given of any validation process, other than simple assessments of test-retest reliability and face validity. Eleven questions were asked on aspects of family life disruption, and five on the child's own HRQOL. The instrument did not detect any significant HRQOL impairment for families overall. For the families of severely affected children who required surgery, however, there was a significantly greater impact compared to the families of children managed expectantly.

The CHQ includes questions in four of its domains which come under the heading of quality of family life. While this makes it one of the few generic instruments to assess family impact, the benefit of this is lost when the child- and family-centred information is combined in the two summary scores, Physical and Psychosocial.

3.5 Comment

The children seen by otolaryngologists have certain unusual characteristics that are important when considering HRQOL assessment: most notable are young age and a high prevalence of problems affecting communication. A large number of tools for assessing condition-specific handicap are emerging, but experience with generic instruments is limited. Quality of family life has not been adequately addressed, and a generic instrument specifically worded to assess benefit after an intervention in children is lacking.

4 Study overview

Otolaryngological conditions, such as otitis media with effusion, are still the commonest reasons for children in the UK to be subjected to a general anaesthetic. Otolaryngologists are under increasing pressure from the purchasers of health care to provide evidence that their treatments are effective. Research with broad outcome measures is required.

It should be obvious from the previous chapters, however, that outcomes research can be a complex and confusing area. There has recently been a proliferation of reports of new HRQOL and condition-specific handicap measures which are described once, and never heard of or seen again. The busy clinician who wishes to investigate the use of outcome measures for audit or research purposes will find the choice overwhelming and the complexity of the issues daunting.

It would seem helpful, therefore, to review the whole field to establish what measures are available and what their merits are. New measures should only be developed to address areas not adequately covered at present. These new measures, properly validated for use in paediatric otolaryngology, can then be presented alongside existing measures as a "clinician's guide to HRQOL assessment".

To achieve this, a large cohort of children attending otolaryngology outpatients is required, offering the opportunity to assess the use of a range of existing generic HRQOL measures in the context of otolaryngology and in a UK population. It was hoped from the outset that at least one of the existing measures would be found to be sufficiently useful as to make the development of a new instrument unnecessary. However, there are two specific areas not adequately covered by any instruments at present, and where new instruments would clearly be required. The assessment of HRQOL benefit from interventions in children is an area not currently well-served, and the development of a Glasgow Children's Benefit Inventory is the first proposal. Quality of Family Life is another area of need, and studies to assess the appropriateness of the MRC Quality of Family Life questionnaire (developed to the initial pilot stage by H Fortnum and others, but never taken further) are the second proposal.

The result will be to provide the otolaryngologist with a range of measures (some new, some established) for use in different circumstances, and with the data available upon which to make an informed choice. These studies form the basis of this thesis and are described in the following chapters.

5 The usefulness of existing generic HRQOL measures for paediatric otolaryngology

5.1 Background

Recurrent acute sore throat, recurrent acute otitis media (RAOM) and otitis media with effusion (OME) are among the commonest diseases of childhood in the developed world. They cause a significant burden of ill health, parental concern, and health care expenditure. Because these conditions are rarely fatal, their impact, and the benefits of their treatment, must be measured in terms of well-being, functional status and quality of life.

Quality of life is a subjective experience, and so can only be assessed by means of questionnaires or interviews. There is no need to produce and validate a new instrument if an adequate instrument exists already. A large number of instruments have been described, with varying amounts of validation data (see Chapter 2). None can yet be considered a "gold standard" although some, such as the CHQ, TACQOL and HUI, have begun to find widespread use. Instruments designed (usually) by paediatricians may perform well in the context of the chronic, stable conditions that they are used to dealing with (asthma, arthritis, skin complaints, etc) but may not necessarily perform well in the context of either common ear and throat infections, which are intermittently severe but with long periods of normality between episodes, or disorders affecting communication (hearing and speech). It is essential to know which

instruments are most suitable for use in otolaryngology to inform the choice of instrument for future otolaryngology research.

5.2 Study aims

The aim of this study was to identify the generic HRQOL measures which have been produced for use in children and which seem to be the best candidates for use in the paediatric otolaryngology population. These instruments would then be applied to a series of children attending otolaryngology clinics with common complaints in order to establish aspects of validity in this context, and ultimately to identify the most suitable instruments for use in future research. Should none prove suitable, a new instrument would need to be produced.

5.3 Participants and methods

5.3.1 Instrument selection

The first step was to decide which instruments to study from the choice available. This was done after a thorough literature review, by choosing those instruments which satisfy the following criteria:

1. Generic measure of HRQOL with published evidence of validity when used in a paediatric population, and of reliability
2. Designed for use in children
3. No trained interviewer required, for reasons of practicality and future widespread applicability
4. Child-completed
5. Suitable for use in pre-school children, since this is the age-group most often affected by common otolaryngological conditions
6. Domain structure includes physical, psychological and social areas
7. Includes assessment of both functional status and subjective well-being

It was found that items 4 and 5 were mutually exclusive for the available range of instruments. For children below the age of 8, no means exist for children to report accurately their own perceived quality of life, due to limitations in vocabulary and abstract reasoning. Parents, therefore, must be used as proxy respondents. Parental reports of their child's quality of life show high levels of agreement with child self-reports where these can be obtained, although more so for easily observable aspects of physical functioning than for social and emotional issues [42].

The HUI was chosen for inclusion in the study, despite the fact that it does not cover the social domain, because it is so widely known and used among health economists (although not as widely used in the UK as the EuroQuol and SF36), and therefore, has much to offer otolaryngologists as a potentially useful health

utility measure. TACQOL/TAPQOL and the CHQ-PF50 were also chosen on the basis that they are the generic instruments which have been studied the most out of the choice available. The CHQ in particular is rapidly becoming the benchmark against which other instruments are compared Given the high proportion of pre-school children seen in otolaryngology clinics, the age range of TAPQOL (1-5 years) was thought to be particularly advantageous, with TACQOL as an equivalent instrument for older children. KINDL and PedsQL were also seriously considered, but neither had been widely used at the time the study was commenced and it was never going to be possible to study more than a handful of instruments at once.

5.3.2 *Patient selection*

The next step was to decide which children to study. Otitis media with effusion, recurrent acute otitis media and recurrent acute sore throat were chosen for study on the basis that they are common, are likely to impact on HRQOL and are associated with professional and public concerns regarding the effectiveness of available treatments.

The children referred to hospital outpatient clinics with these clinical problems, regardless of the ultimate specialist diagnosis, formed the study population. They were found to encompass a wide range of disease severities, from children who were essentially normal and required only parental reassurance, to those who were severely affected and required surgical management.

5.3.3 *Study hypotheses*

The range of disease severity present in the study population allowed us to assess the construct validity of the instruments in the context of paediatric otolaryngology by setting up and testing the following *a priori* hypotheses:

1. The generic instruments should show convergent validity between HRQOL scores and markers of disease severity.
2. The generic instruments should show concurrent validity with disease-specific measures of handicap, such as OM6.
3. The generic instruments should show concurrent validity with each other for similar domains.
4. The instruments should produce results which are independent of a direct influence of age, sex and socio-economic deprivation.

To expand on some of these points, we postulated that there would be a significant impact on HRQOL as a result of the otolaryngological conditions under study. Our hypothesis was that, in general, the magnitude of the impairment in HRQOL would be related in some way to the severity of the disease. We would therefore compare the results for each generic HRQOL instrument with markers of disease severity. For the children with sore throats, these were frequency of sore throat and pyrexia and need to take time off school. For children with recurrent acute otitis media these were frequency of

otalgia and pyrexia, and days lost from school. For children with otitis media with effusion, these were pure tone thresholds, tympanometry results and presence of concerns regarding speech development.

In general, the generic HRQOL instruments are attempting to measure the same things, and it should therefore be possible to demonstrate concurrent validity for similar domains. Specifically, these are the domains relating to emotional and psychological issues in the three instruments; the domains relating to social issues in the CHQ and TACQOL; and the domains relating to physical issues including pain in the three instruments.

5.3.4 Method of evaluating instrument validity

Prior approval for the study was obtained from local research ethics committees and written consent for study participation was obtained for every child.

A consecutive series of children aged 1-16 years was recruited for the study from the paediatric otolaryngology clinics of three hospitals in the West of Scotland (Crosshouse Hospital, Kilmarnock; Ayr Hospital; The Royal Hospital for Sick Children, Yorkhill, Glasgow). To be eligible for inclusion, the children had to be at their first hospital visit after being referred by their General Practitioner with suspected otitis media with effusion (OME), recurrent acute otitis media (AOM) or recurrent sore throats.

At the time of their hospital visit, the parents were asked to complete a range of HRQOL measures on behalf of the child. All were asked to complete the HUI mark III together with a global rating of the child's HRQOL on a 10cm visual analogue scale. For those presenting with OME or recurrent AOM, the otitis media-related handicap measure OM6 [7, 8] was also completed. Due to age restrictions in the design of the instruments, the Child Health Questionnaire (50-item parent-completed version) [6] was given only to parents of children aged 5 years and above. The TAPQOL was given to parents of children aged 1-5 years and TACQOL to parents of those aged 6 years and above. The order in which the instruments were presented was varied randomly. All children then underwent a standard clinical consultation with the same otolaryngologist (HK) where clinical data were collected.

Data were stored on a computer, and statistical analyses were performed using SPSS version 11.0.

5.4 Results

5.4.1 The study cohort

274 children were seen in the clinics. Twenty two declined to participate in this part of the study, leaving 252 children for analysis. 130 were boys, 122 girls, and the median age was 5 (range 1-14 years). The primary referral diagnosis was OME in 124 cases, recurrent AOM in 58 and sore throats in 70. In many cases, however, more than one symptom was present on enquiry. A study recruitment flow diagram is shown in Appendix 2

5.4.2 The Health Utilities Index

Completed questionnaires were obtained for all 252 children. A wide range of scores was obtained for the single- and multi-attribute utilities. The ranges, means and standard deviations for the study group as a whole are shown in Table 5.1.

Multi-attribute and single-attribute utilities did not vary in any consistent way with the sex of the child, the sex of the person filling in the form, socioeconomic deprivation of area of residence (assessed by the Carstairs Deprivation Index [9]), or occupation of parents (manual versus non-manual). Table 5.2 shows the statistical significance of the associations between single and multi attribute utilities and these variables. Some associations are shown, but these are

assumed to be spurious given the large number of statistical analyses performed and the lack of any *a priori* hypotheses to support such associations.

Although the HUI mark III has a stated lower age limit of 6 years, the lack of an alternative health utility measure for use in pre-school children led us to test this lower age limit and determine the lowest age at which the HUI still performs adequately. Children aged 3 years or less had poorer multi-attribute utilities than older children (Jonckheere-Terpstra test*, p=0.033), with the difference being due to much poorer scores for speech (p<0.001, Figure 5.1) and ambulation (p<0.001). Scores for the other single-attribute utilities were not affected by age (Table 5.2).

Although there was no difference in the multi-attribute utilities between children with a referral diagnosis of OME, recurrent AOM or sore throats (Kruskal-Wallis, p=0.172), there were differences between these groups in the single-attribute utilities. The hearing and cognition utility scores were worse in the OME group (p=0.001 for each, Figure 5.3); speech utility scores were worse for those with either OME or recurrent AOM (p<0.001, Figure 5.3); and pain utility scores were worse in those with either recurrent AOM or sore throats (p<0.001, Figure 5.2).

* The data are in the form of a continuous variable (HUI score) which is not normally distributed in this sample. Non-parametric tests are therefore appropriate. Where the median values of 2 groups are being compared, this would be a Mann-Whitney U test, and for more than 2 groups the equivalent test would be the Kruskal-Wallis test. However, in this case, the groups being compared have a natural order (5 reported levels of parental satisfaction) and the Kruskal-Wallis test does not take this into account. The appropriate test in this situation is the Jonckheere-Terpstra test, which is a non-parametric comparison of medians between a series of ordered groups.

For the children with recurrent pyrexial illness (AOM or sore throats), the average number of days with pyrexia per month was weakly correlated with the multi-attribute utility (Spearman's rho=-0.174, p=0.046, Figure 5.4) and with the single-attribute utilities for pain (rho=-0.337, p<0.001) and emotion (rho=-0.260, p=0.001). Utilities were not, however, worse in those who had needed time off school compared with those who had not.

In the children with sore throats, the number of sore throats in the last year was inversely correlated with the multi-attribute utility (rho=-0.378, p=0.002, Figure 5.5) and the single-attribute utilities for pain (rho=-0.383, p=0.001) and emotion (rho=-0.251, p=0.032). In the children with recurrent AOM, the average number of episodes of otalgia per month was inversely correlated with the pain single-attribute utility (rho=-0.276, p=0.009, Figure 5.6), but not with any other utilities. Frequency of otorrhoea had no effect on utilities.

In the children with OME, hearing single attribute utilities were worse in those who had middle ear fluid confirmed on tympanometry in both ears (type B or C2 tympanograms) compared with those who did not (n= 51 and 52 respectively; Mann-Whitney, p=0.005; Figure 5.7), but the other utilities were not affected. Those children who had a bilateral hearing impairment of at least 25dB had significantly worse multi-attribute utilities than those with better hearing (n=38 and 64 respectively; Mann-Whitney, p=0.017, Figure 5.8). The same was true for the single attribute utilities for hearing and speech in these children (p=0.003 and 0.025 respectively, Figure 5.8).

In the children with OME or recurrent AOM, ear-related handicap rated on the OM6 questionnaire was significantly inversely correlated with the multi-attribute utility (rho=-0.608, p<0.001, Figure 5.9) and the single-attribute utilities for hearing (rho=-0.369, p<0.001), speech (rho=-0.283, p<0.001), emotion (rho=-0.476, p<0.001), pain (-0.611, p<0.001) and cognition (rho=-0.215, p=0.005).

Overall health-related quality of life, rated either on a 10cm visual analogue scale or on a 5-point Likert-type rating scale, was significantly associated with the multi-attribute utility (Spearman's rho=-0.408, p<0.001, Figure 5.10; and Jonckheere-Terpstra, p<0.001, Figure 5.11, respectively) and with the single-attribute utilities for hearing, speech, emotion, pain and cognition.

5.4.3 The Child Health Questionnaire

Completed questionnaires were obtained for 109 children, of whom 55 were girls and 54 boys. They ranged in age from 5 to 14 years with a median of 7 years and a mean of 7.5. The primary reason for referral was OME in 58, recurrent AOM in 13 and sore throats in 38.

The 50 items in the CHQ showed a high degree of internal consistency with a Cronbach's alpha of 0.783. There was, however, evidence of significant ceiling effects in the following domains: Physical Functioning, Role/Social Limitations

Physical, Role/Social Limitations Emotional, Bodily Pain, Self Esteem, Parent Impact Emotion, Parent Impact Time and Family Activities. In each of these domains, the modal response was the maximum possible value, accounting for 20-75% of all responses (Figure 5.12).

The use of two summary scores and the weightings used to calculate them were originally based on a factor analysis of ten of the domain scores of the CHQ-PF50. An attempt was made to replicate the factor analysis using the data from this study. Analysis was performed by extracting principle components with varimax rotation and selecting for eigenvalues greater than 1, as reported by the originators of the CHQ [56]. Only two factors were extracted which between them accounted for 61% of the variance. The degree to which each domain score loaded onto the two factors differed in some respects from the US validation data, however, as shown in Table 5.3. Factor loadings for Physical Functioning, Role/Social Limitations Physical, Self Esteem, General Health Perceptions, Parent Impact Time and Parent Impact Emotion were all very similar to the USA data. Significant differences were observed for Behaviour, Role/Social Limitations Emotional and Mental Health, which all loaded onto the Physical factor in this study rather than the Psychosocial factor as in the USA, and Bodily Pain, which loaded onto the Psychosocial factor rather than the Physical one as in the USA.

The CHQ domain and summary scores in the study sample are shown in Table 5.4. There was no association between any of the following factors and either

the Physical or the Psychosocial summary score: age of the child (Jonckheere Terpstra, p=0.745 and p=0.889 respectively), sex of the child (Mann Whitney, p=0.774 and p=0.576), sex of the parent completing the forms (Mann-Whitney, p=0.192 and p=0.861), manual or non-manual occupation of the parents (Mann-Whitney, p=0.220 and p=0.265) and socio-economic deprivation estimated using the Carstairs Deprivation Index (Jonckheere Terpstra, p=0.706 and p=0.297).

Children with recurrent AOM showed the greatest impairment, and children with OME the least, in both the Physical and Psychosocial summary scores (Kruskal Wallis, p=0.021 and p=0.032), and also in six of the domain scores (Global Health, p=0.012; Physical Functioning, p<0.001; Role/Social Limitations Physical, p=0.042; Mental Health, p=0.018; General Health Perceptions, p=0.003; Family Activities, p<0.001; Figure 5.13).

In children with either recurrent AOM or sore throats, CHQ summary and domain scores were not different in children who had lost time from school compared with those who had not. However, the average number of days per month with pyrexia was correlated with both the Physical summary score (Spearman's rho=-0.311, p=0.018) and the Psychosocial summary score (rho=-0.394, p=0.002, Figure 5.14). Frequency of pyrexia was also significantly correlated with eleven of the fifteen domain scores (Table 5.5).

For children with sore throats, those with fewer than 6 sore throats per year (the median value) had significantly better scores for the Bodily Pain domain (Mann Whitney, p=0.016) as well as Global Health (p=0.031) and Parent Impact Emotion (p=0.035), but the Physical and Psychosocial summary scores were not significantly different (Figure 5.15).

In children with OME, only the General Health Perceptions domain score was significantly associated with either a better-ear audiometric threshold of 25dB or worse (Mann Whitney, p=0.008) or the presence of bilateral type B/C2 tympanometry (p=0.013, Figure 5.16). In children with recurrent AOM, frequency of otalgia and frequency of otorrhoea showed no association with any of the CHQ domains or summary scores. In those children with OME or recurrent AOM, both the Physical and Psychosocial summary scores were correlated with ear-related handicap rated using OM6 (Spearman's rho=-0.373, p=0.002 and rho=-0.404, p=0.001 respectively, Figure 5.17).

Considering the entire study group together, the child's overall HRQOL rated on a 100mm visual analogue scale was more highly correlated with the Physical than the Psychosocial summary score (Spearman's rho=-0.410, p<0.001 and rho=-0.226, p=0.026 respectively, Figure 5.18). Both summary scores were associated with the child's overall HRQOL rated on a 5-point Likert rating scale (Jonckheere Terpstra, p<0.001 in both cases, Figure 5.19).

The two summary scores (Physical and Psychosocial) were significantly associated with the HUI mark III multi-attribute utility (rho=0.498 and 0.499 respectively, $p<0.001$) and with the single-attribute utilities for emotion, pain, dexterity and cognition. Of the 15 domains of the CHQ, most do not have any obvious similarity to the single-attribute utilities of the HUI. There was, however, a high degree of correlation between the HUI single attribute utility for pain, and the Bodily Pain domain score of the CHQ (Spearman's rho=0.725, $p<0.001$). Spearman's correlation coefficients between 0.4 and 0.5 were obtained between the pain single attribute utility and three other domains of the CHQ (Global Health, Role/Social Limitations-Physical, and Family Activities). The only other correlations with Spearman's coefficients of this magnitude were between the emotion single attribute utility and 7 of the CHQ domains (Role/Social Limitations-Physical, Bodily Pain, Mental Health, Self Esteem, Parental Impact-Time, Parental Impact-Emotional, Family Activities).

5.4.4 TACQOL

Completed questionnaires were obtained for 74 children, of whom 36 were girls and 38 boys. They ranged in age from 6 to 14 years with a median of 8 years and a mean of 8.7. The primary reason for referral was OME in 41, recurrent AOM in 6 and sore throats in 27.

TACQOL produces no overall summary score, but rather a series of seven domain scores: Body, Motor, Cognition, Autonomy, Social, Positive Emotions

and Negative Emotions. Parents are asked to respond with reference to the child's problems over the last 3 months. For items in the first five of these domains, the parent gives separate scores for the presence of the problem, and, if present, the child's emotional reaction to it. The Positive Emotions and Negative Emotions sub-scales do not have any such additional rating. Scores for each sub-scale are calculated by simple addition of question scores, giving scores from 0-32 for the 5 emotionally-weighted sub-scales, and scores of 0-16 for the Positive Emotions and Negative Emotions sub-scales. No overall score is calculated. Domain scores in our study sample are given in Table 5.6.

The 57 items in the TACQOL (not including the qualifying statements regarding emotional response) showed a high degree of internal consistency, with a Cronbach's alpha of 0.893. The internal consistency of each of the domain scores ranged from 0.637 to 0.921 (Table 5.7).

There was evidence of a ceiling effect in all domains except Negative Emotions and Body. For the other five domains, the maximum possible score (16 for Positive Emotions, 32 for all others) was the modal score and accounted for 20-72% of all responses. For Body, the modal score was 28 and scores were well-distributed across the possible range, although skewed towards the top of the range. Scores for Negative Emotions were quite evenly distributed across the range with a mode of 10 (Figure 5.20).

There was no consistent association between TACQOL domain scores and age of child, sex of child, sex of respondent (mother versus father), occupation of main wage-earner (manual versus non-manual) or the degree of socio-economic deprivation of their area of residence (estimated with the Carstairs Deprivation Index [111]), as shown in Table 5.8. One domain score did show a statistically significant association (Carstairs Deprivation Index and Motor), but the association is weak and, in the absence of an *a priori* hypothesis to support such an association, it is assumed to be spurious. The Motor domain also showed an association with age, and this could possibly have been a genuine effect with the youngest children in the sample (aged 6 years) being unable to perform some of the motor tasks listed. Examination of the data, however, shows no clear trend towards worse scores in the youngest children, and the association depends entirely on the presence of a small number of extreme outliers.

Domain scores appeared to be worse in children with recurrent AOM compared to those with OME or sore throats, but the number of children in this group is very small. The difference was statistically significant for Body, Motor and Autonomy (Kruskal Wallis, p=0.015, 0.02 and 0.006 respectively, Figure 5.21).

For the children with recurrent pyrexial illness (AOM or sore throats), there was a significant correlation between the average number of days pyrexia per month and the domain scores for Body, Motor, Autonomy and Positive Emotions (Spearman's rho=-0.327, -0.332, -0.590 and -0.340 repectively, p=0.045, 0.039, <0.001 and 0.034, Figure 5.22). There was no association between domain

scores and the need to take time off school (Mann Whitney, $p>0.05$ for all seven domains).

For the children with recurrent sore throats, there was no significant correlation between the number of sore throats in the last 12 months and the domain scores, although for at least two domains this is likely to be due to inadequate numbers (Body domain, Spearman's rho=-0.369, p=0.058; Motor domain, rho=-0.359, p=0.060; Figure 5.23).

For children with OME, the Body domain score was associated with the presence of type B/C2 tympanometry (Mann Whitney, p=0.035, Figure 5.24), but there was no association with any of the other domains. None of the domains were associated with the presence of a better-ear threshold of 25dB or worse. Ear-related handicap rated using OM6 was correlated with five of the domain scores; Body, Motor, Autonomy, Cognition and Social (Spearman's rho=-0.569, -0.402, -0.315, -0.401 and -0.416 respectively, p= <0.001, 0.006, 0.031, 0.005 and 0.004, Figure 5.25).

All domain scores were correlated with overall HRQOL rated on a 100mm visual analogue scale and on a 5-point Likert rating scale, as shown in Table 5.9 and Figure 5.26.

5.4.5 TAPQOL

150 children were recruited to this part of the study, comprising 76 boys and 74 girls, aged between 1 and 5 years (mean 3.4, median 4). The primary reason for referral to hospital was OME in 67, recurrent AOM in 47 and recurrent sore throats in 36.

The 46 items in the TAPQOL (not including the qualifying statements regarding emotional response) showed a high degree of internal consistency, with a Cronbach's alpha of 0.821. The internal consistencyof the items in each domain ranged from 0.419 to 0.949 (Table 5.10).

TAPQOL produces no overall summary score, but rather a series of domain scores, each scored between 0 (worst quality of life) and 100 (best quality of life). The published version of TAPQOL has 43 questions in 12 domains [70]. The English language version supplied by the originators for our study (which began before publication of the article above) contains 46 questions in 13 domains. The domains are sleep, appetite, respiratory problems, abdominal problems, skin problems, motor function, social behaviour, communication, positive mood, anxiety, aggression, eating problems and vitality. Of these, the domains for motor function, communication and social behaviour are only answered by parents of children aged 18 months or older, as the questions are not suitable for younger children. There are between 3 and 7 items within each domain. Parents are asked to respond with reference to the child's problems

over the last 3 months. For each item, the parent gives separate scores for the presence of the problem, and, if present, the child's emotional reaction to it.

TAPQOL scores showed a ceiling effect for most of the domains, with the most common score being 100 (the maximum possible). The exceptions to this were aggression (modal value 57) and sleep (two modal values, 50 and 75). In the motor domain no child scored below 75, and in the eating problems and positive mood domains no child scored below 50. For the remainder, a wide range of scores were obtained, and there were even some zero scores (the minimum possible) in the domains of communication, vitality, aggression, sleep and skin problems. An example of a domain with a ceiling effect is given in figure 5.27.

There was no consistent association between TAPQOL domain scores and age of child, sex of child, sex of respondent (mother versus father), occupation of main wage-earner (manual versus non-manual) or the degree of socio-economic deprivation of their area of residence (estimated with the Carstairs Deprivation Index [111]), as shown in Table 5.11. There were three domain scores which did show a statistically significant association (age of child and Vitality, sex of respondent and Aggression, sex of child and Motor Functioning), but with such a large number of statistical analyses being performed, some spurious associations are to be expected and apparently significant p values should be treated with caution in the absence of an *a priori* hypothesis to support an association.

Domain scores for appetite, eating problems and vitality were all worse in the children with sore throats, compared to those with OME or recurrent AOM (Kruskal Wallis, $p<0.001$; Figure 5.28). Scores for the communication domain were slightly worse in the OME group (Kruskal Wallis, $p=0.014$). Scores in the sleep domain were worse in the children with pyrexial illnesses (recurrent AOM or sore throats) than the children with OME (Kruskal Wallis, $p=0.006$). No other effect of diagnosis on TAPQOL scores was seen. Domain scores for each group are given in Table 5.12.

For the children with recurrent pyrexial illness (AOM or sore throats), there was a significant correlation between the average number of days pyrexia per month and 7 of the 13 domains, as shown in Table 5.13. An example is shown in Figure 5.29. The domain scores for eating problems and vitality were significantly worse in those children who had needed to take time off school than in those who had not (Mann Whitney, $p=0.01$ and $p<0.001$ respectively). For the children with sore throats, the number of sore throats in the last year was significantly correlated with 8 of the 13 domain scores, as shown in Table 5.14.

For the children with OME, there was no association between the TAPQOL domain scores and either the presence of bilateral type B or C2 tympanograms, or a better ear threshold of better than 25dB (Figure 5.30). For the children with recurrent AOM, the domain scores did not correlate with frequency of otalgia

or otorrhoea. For the children with OME or recurrent AOM, otitis media-related handicap was assessed with the OM6 questionnaire. OM6 scores were correlated with 9 of the 13 TAPQOL domain scores, as shown in Table 5.15. An example is shown in Figure 5.31.

The child's overall health-related quality of life, as rated by parents on a 10cm visual analogue scale, was significantly correlated with only 4 domains: sleep (rho=-0.321, p=0.001), positive mood (rho=-0.306, p=0.001), appetite (rho=-0.274, p=0.004) and motor functioning (rho=-0.262, p=0.009). There was a significant association between overall health-related quality of life rated on a five-point Likert rating scale, however, and 9 of the 13 TAPQOL domain scores: sleep, appetite, eating problems, vitality, positive mood, aggression, social behaviour, motor functioning and communication (Jonckheere-Terpstra test, $p<0.05$ – Figure 5.32).

TAPQOL showed a degree of convergent validity with the HUI. TAPQOL domain scores were all correlated with the HUI mark III multi attribute utility (Table 5.16 and Figure 5.33). The sleep domain was most highly correlated with the pain single attribute utility (Spearman's rho=0.559, $p<0.001$). The appetite domain was correlated with the emotion and pain utilities (rho=0.302 and 0.481 respectively, $p<0.001$ for both). The eating problems domain was most highly correlated with the pain utility (rho=0.421, $p<0.001$). The positive emotions domain was correlated with utilities for emotion and pain (rho=0.373 and 0.318, $p<0.001$). The aggression domain was most highly correlated with the emotion

utility (rho=0.302, p<0.001). The social behaviour domain was correlated with the utilities for hearing and cognition (rho=0.309 and 0.317, p<0.001). The communication domain was highly correlated with the utilities for hearing, speech and cognition (rho=0.345, 0.604 and 0.416, all p<0.001).

5.5 Discussion

5.5.1 *The Health Utilities Index mark III*

The limited funding available in a publicly-funded health service such as that in the UK makes economic evaluations increasingly important. The ability to assess not only the health gains of an intervention, but also the cost of those gains, is highly desirable when competing for scarce resources within the health service.

The HUI is widely known and used by health economists for determining health utilities, which can be used to calculate the familiar "cost per quality-adjusted life year". Before we allow such calculations to be performed in paediatric otolaryngology, we must ensure that the instruments being used are adequate for their purpose. We know that the HUI is not sensitive to the impairments present in children with asthma [3], for example, so it is important to know the extent to which the HUI is sensitive to the impairments present in children with common ear and throat conditions.

The children in the study were an unselected series of referrals to hospital, and therefore represented a wide range of disease severity, ranging from severely affected children who required surgery to children hardly outside the normal range of experience, who simply required reassurance and explanation. As a result of this, a wide range of HUI scores was produced (Table 5.1). For some of

the single attributes, such as vision, the scores clustered at the higher end of the scale with little spread. For others, however, there was a large spread, including some children who had health utilities of zero for some attributes. This is a health state rated by the normal population as being as bad as death were it to be permanent. It may seem surprising that parents regard ear infections and sore throats as being of this severity, when many health professionals would regard these conditions as being relatively trivial. Clearly, not all parents rated the problems as being this severe, but it is precisely because diseases produce such an unpredictable impact on people that health-related quality of life is worth measuring at all.

Although it is reassuring to see that HUI scores are not unduly influenced by sex and social class, there is a lower age limit beyond which the HUI is not useful. This is because the questions in the HUI are not referenced to what would be expected for a normal child of the same age. This is particularly important for vision ("can read newsprint"), speech ("can be understood by strangers"), mobility ("can walk without assistance") and self-care ("can eat, dress, bathe and use the toilet without assistance"), where very young children would not ususally be able to perform these functions unaided. This is potentially a problem in paediatric otolaryngology, where many of our common diseases are most prevalent in pre-school children. However, it seems that we could potentially extend the use of the HUI mark III beyond the lower limit of 6 years suggested by the originators down to age 4, based on the data above.

The HUI does seem to be sensitive to the particular impairments present in the children studied. As one would expect, children with OME scored poorly for hearing and speech single attribute utilities, whereas children with recurrent ear or throat infections scored poorly on the pain single attribute utility. Frequency of pyrexial illness, sore throat and otalgia were all associated with poorer scores for the pain single attribute utility, again as one would expect. Tympanometry and audiology findings were also reflected in the hearing single attribute utility.

HUI utility scores were correlated with ear-related handicap, as measured on OM6, and also with ratings of overall health-related quality of life on a 5 point scale and a 100mm visual analogue scale. Interestingly, the HUI scores were also correlated with the CHQ summary scores and TAPQOL domain scores, despite completely different underlying philosophies of the instruments (physical functioning almost exclusively for the HUI versus mutli-dimensional, including psychosocial, for the CHQ and TAPQOL) and scoring methods (population preference-based utility versus summation of rating scales). This is all supporting evidence for the validity of the HUI as a simple measure which can genuinely reflect the broad nature of health-related quality of life. The degree to which the single attribute utilities and the CHQ subscales correlate with each other is largely predictable, and in keeping with a previous comparison of the two instruments [10]. The association between the HUI and

TAPQOL has not been reported before, but the pattern of associations is also largely in keeping with what one would expect.

In summary, it seems that the HUI mark III provides a useful measure of health-related quality of life in children with OME, recurrent AOM and sore throats, as long as its use is restricted to children at least 4 years of age. The HUI is sensitive to the nature and degree of impairments present in these children. It should be useful in future economic evaluations.

5.5.2 *The Child Health Questionnaire*

The CHQ is probably the most widely used and known generic HRQOL measure designed for use in children. It is limited to children aged at least 5 years old, which limits its usefulness in otolaryngology, as shown by the fact that data from the CHQ could only be collected on 43% of the children recruited to the study overall.

The presence of significant ceiling effects in some domains may reduce the sensitivity of the CHQ and this needs to be specifically addressed in future studies. However, it may be that these ceiling effects are only a feature in this particular study population, for whom the areas concerned are not relevant. They may well be more relevant in other areas of paediatric medicine, and ceiling effects may not be apparent in other studies.

A study of normal Australian children showed that the factor structure of the CHQ-PF50 that is used as the basis for the two summary scores could not be replicated [112]. However, the factor analysis reported here does support the two-factor model underlying the summary scores. There are some differences in the factor loadings that make the labelling of the factors as "Physical" and "Psychosocial" less convincing, but overall they are probably still broadly in keeping with the model proposed by the originators of the CHQ [56]. In fact, the factor loadings reported here are probably closer to the USA data than those from a previous attempt to replicate the CHQ factor structure in a UK otitis media population [38].

The CHQ manual [56] contains data on a number of reference samples of healthy and diseased children that can be used for comparison with the data presented in Table 5.4. The CHQ domain and summary scores were all poorer in this study sample than in the published data on normal children, with the exception of the Self Esteem and Family Cohesion domains. The domains for Physical Functioning, Role/Social Limitations Physical, General Health Perceptions and BodilyPain and the Physical summary score were all poorer than in a reference sample with attention deficit – hyperactivity disorder. The domain scores and Psychosocial summary scores were poorer than scores in some if not all of the four published reference samples with asthma. The domain scores for Role/Social Limitations Physical, Role/Social Limitations Emotional, Parent Impact Time, Self Esteem, Mental Health and Behaviour and the Psychosocial summary score were all poorer than in the reference sample with juvenile

chronic arthritis. Only the domain scores for Bodily Pain, Mental Health, Behaviour, Family Cohesion and Role/Social Limitations Emotional were poorer than in a reference sample with epilepsy.

The data here showed no influence on CHQ scores of extraneous variables such as age, sex or socio-economic deprivation, a fact which supports the robustness of the CHQ. This is in contrast to data from the validation studies in the USA which do show significant effects of all these variables, as well as ethnicity [56]. The originators seem to regard this as an acceptable feature of the instrument.

In this study, the Psychosocial summary score was found to vary with disease severity almost as much as the Physical summary score. This is in contrast to previous studies using CHQ-PF28 in children with sinus disease and sleep disordered breathing [84, 88], where only the Physical summary score was associated with improvement after surgery. This may reflect the greater sensitivity of the 50-question version compared to the 28-question short form.

The CHQ summary scores and domains showed a reasonable degree of association with markers of disease severity such as frequency of pyrexia and sore throat. Correlation with ear-related handicap was better than physical measures such as frequency of otalgia and hearing thresholds. In fact, the CHQ appears to be largely insensitive to the impairments present in children with OME, which may explain why children with OME showed less impairment of HRQOL overall than children with recurrent AOM or sore throats. This is not

surprising when the range of items in the CHQ is studied: communication issues do not feature at all. This may prove to be a major issue for the applicability of the CHQ to otolaryngology populations.

However, the CHQ summary scores correlated well with direct ratings of HRQOL, and the performance of the instrument overall was reasonable. With regard to its face validity, its items cover a broad range of areas of the child's day-to-day life without resorting to a list of diseases or symptoms, and it is, therefore, a genuinely promising generic HRQOL measure. Its use in American sudies is now widespread and it is rapidly becoming the "gold standard" in its field. We will, no doubt, see more use of it in otolaryngology research.

5.5.3 TACQOL

The main reason for studying TACQOL here was as a counterpart to TAPQOL for older children. The most obvious conclusion to emerge is that TACQOL's age-range is applicable to only a small proportion of the paediatric otolaryngology outpatient population (29% in this study), and small numbers clearly limit what further conclusions can be drawn. Discussion here will be limited as a result, and also because much that can be said about TACQOL will appear in the discussion about TAPQOL.

Although some associations were found with markers of disease severity such as frequency of pyrexia and tympanometry, many associations could not be reliably assessed due to small numbers. However, correlation with ear-related handicap and overall HRQOL was good, and other associations may emerge if an adequately-powered study were performed.

It is, perhaps, unsurprising that the Body domain showed a reasonable degree of association with markers of disease severity given that the first question it contains asks specifically about frequency of ear infections and sore throats. In fact, it is the only generic HRQOL measure to do so. For this reason alone, the instrument deserves study for use in otolaryngology.

The Body domain scores differed markedly from the normal reference data published in the TACQOL manual [24]. The mean score in our data was 5 points lower than the mean in the normal sample for this domain (Table 5.6); other domains differed by only a point or so. However, the scores for all the domains were still lower than those for children with chronic conditions (asthma, epilepsy, arthritis, allergies, diabetes and heart conditions), those on medical treatment and those with recent upper respiratory tract infections [24].

One problem with TACQOL to emerge from our data is the ceiling effect in many of the domains. Since all children cluster near the top of the range of possible scores, the domain scores may lack sensitivity, such that it may be difficult to show changes with treatment. This issue needs to be specifically

addressed in future studies. It remains to be seen whether these ceiling effects are a feature peculiar to this study population and are simply a reflection of the specific range of areas affected by the conditions under study.

TACQOL shows promise for use in an older otolaryngology clinic population, but larger studies are needed.

5.5.4 TAPQOL

TAPQOL has the potential to be very useful, in that it is one of the few instruments currently available that is specifically designed for use in pre-school children: children of this age are developing rapidly, and questionnaires designed for older children may not address the most appropriate issues.

One strength of TAPQOL, like TACQOL, is that it is the extent to which a problem causes emotional disturbance in the child that determines much of the score, rather than just the presence of the problem, or the concern felt by parents. Although it may appear at first that many of its questions (at least the first 9) are just descriptions of symptoms, rather than attempts to determine how the symptoms affect the child's day to day life, the symptom responses are modified according to the impact they have on the child, such that the outcome is indeed within the scope of HRQOL.

Another strength is that its questions are clearly appropriate for the intended age group. Its potential drawback, however, is that it has an upper age limit of 5 years, which may limit its usefulness in following children up over time.

The originators of TAPQOL have very deliberately refused to produce an overall summary score of any kind, on the grounds that there is no theoretical justification to summate the effects of a disease on very different areas of a person's life [24]. They prefer to think of quality of life as a multi-faceted concept, and to report it solely in terms of domain scores. This makes the results somewhat cumbersome to report and use. It also necessitates multiple statistical comparisons when the data are analysed, and allowance must be made for this with cautious interpretation of any statistically significant associations.

Our results here show that TAPQOL is robust as a measure, without any undue effect of age, sex or socio-economic class. The fact that it can be used to generate meaningful results in children with ear and throat disorders, in addition to the groups of pre-term and chronically ill children studied previously [70], supports its use as a generic measure for children with a wide range of health problems.

Comparison of our results (Table 5.11) with published data [70] show that for virtually all the domain scores, the children in our study had worse scores than children born pre-term or children with chronic diseases (mostly asthma and

bronchitis), and considerably worse than a healthy population sample. Our results show that recurrent AOM, OME and sore throats have a substantial (and measurable) impact on a child's quality of life.

The children in this study are a heterogenous group consisting of an unselected series of children referred to hospital. Some have very mild problems, hardly enough to be considered abnormal, while others have severe problems requiring surgery. This range of severity allows us to test the hypothesis that the children with more severe disease should have worse scores on TAPQOL if it is to be any use as a measure of health-related quality of life. The associations shown between domain scores and markers of disease severity (frequency of pyrexia, sore throat, time off school) largely support this hypothesis, although TAPQOL may lack sensitivity to some of the impairments present in children with OME.

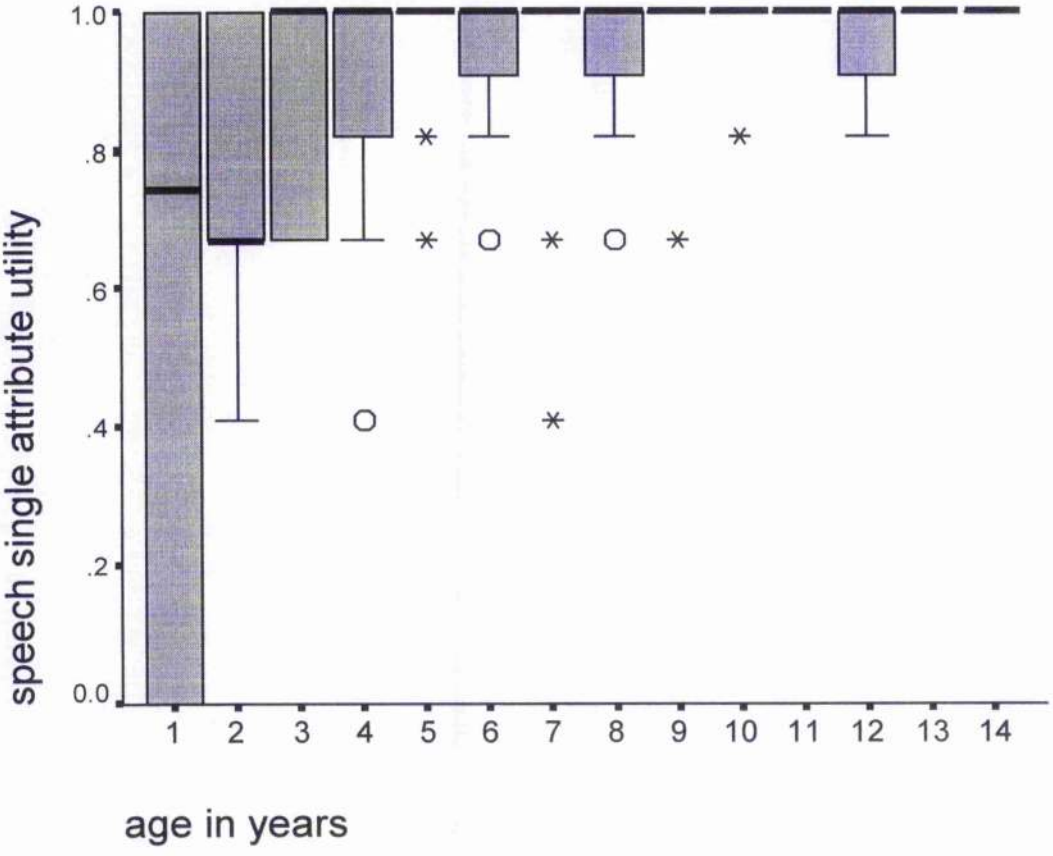
The other hypothesis we wished to test was that TAPQOL scores should correlate to some degree with overall estimates of health-related quality of life, and indeed this seems to be the case for the Likert scale and the visual analogue scale.

The problem of ceiling effects is as much evident with the TAPQOL as with the CHQ and TACQOL as discussed above.

We can conclude that TAPQOL addresses a reasonable range of age-appropriate issues, correlates with disease severity and other measures of quality of life, and is free from any undue influence of age, sex and socio-economic class.

Figure 5.1 Age and HUI speech single attribute utility

The effect of the child's age on scores obtained for the speech single-attribute utility (n=252; Jonckheere-Terpstra test, $p<0.001$) *.



* Much of the data in this thesis is presented as boxplots. In each case, the box shows the 25th, 50th, and 75th centiles, the whiskers the 5th and 95th centiles. A circle represents an outlier between 1.5 and 3 times the interquartile range, an asterisk an extreme outlier more than 3 times the interquartile range.

Figure 5.2 HUI pain single attribute utility & referral diagnosis

The relationship between referral diagnosis and scores for the pain single-attribute utility. Utilities are significantly worse in children with sore throats and recurrent AOM (Kruskal-Wallis, $p<0.001$).

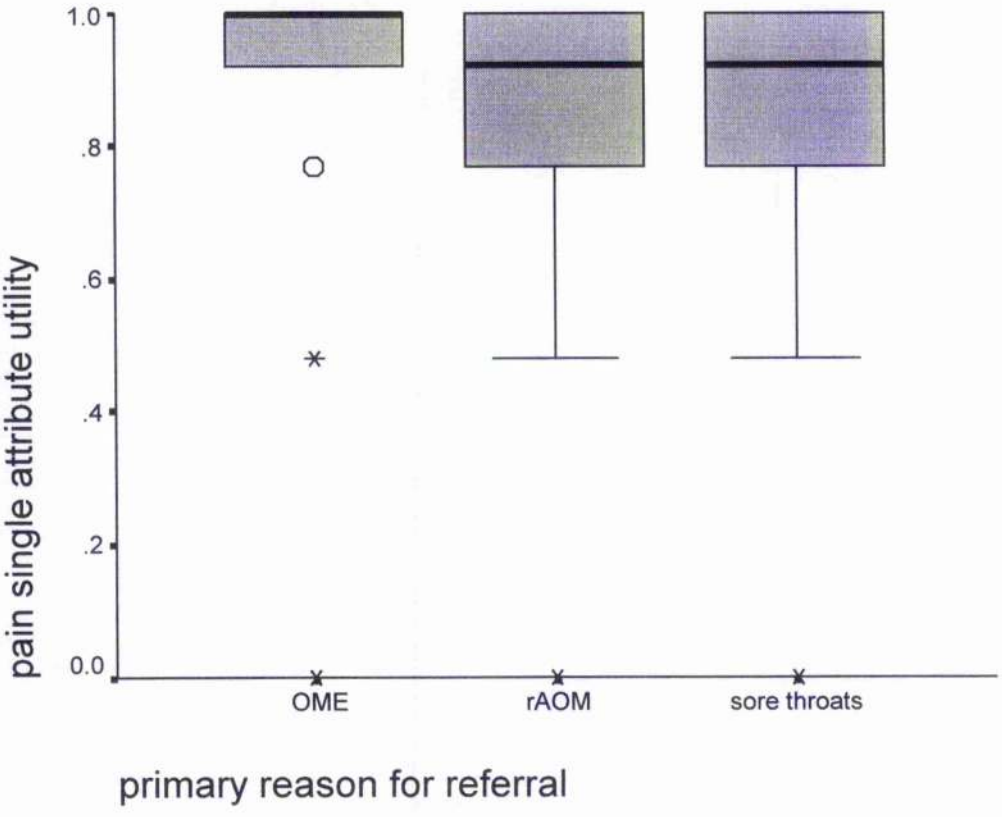
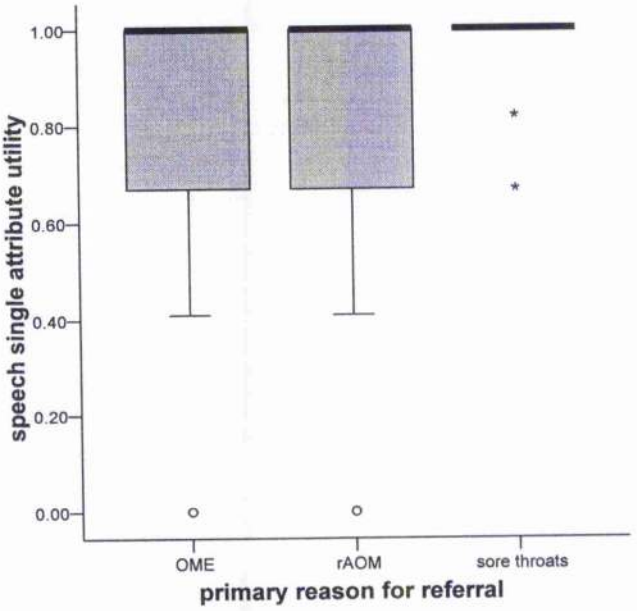


Figure 5.3 HUI speech & cognition utilities & referral diagnosis

The relationship between referral diagnosis and scores for the speech and cognition single-attribute utilities. Speech utilities are significantly worse in children with OME and recurrent AOM (Kruskal-Wallis, $p<0.001$), while cognition utilities are worse in those with OME ($p=0.001$).



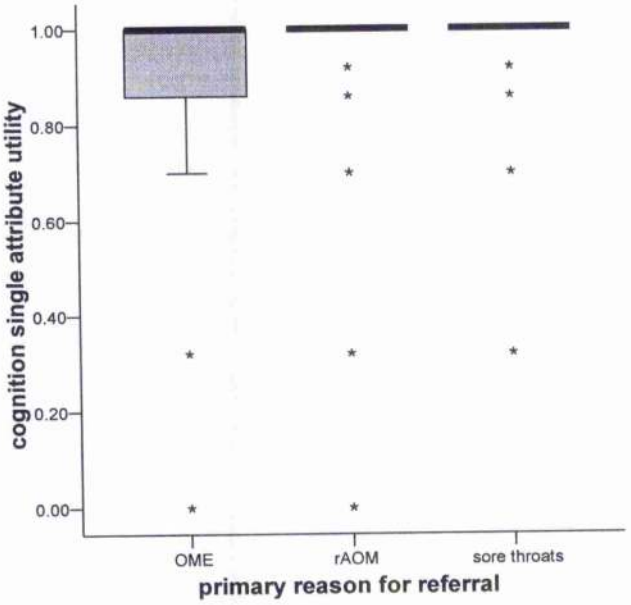


Figure 5.4 HUI multi attribute utility and frequency of pyrexia

There is a weak relationship between multi-attribute utility and frequency of pyrexia in the children with recurrent AOM or sore throats (Spearman's rho=-0.174, p=0.046).

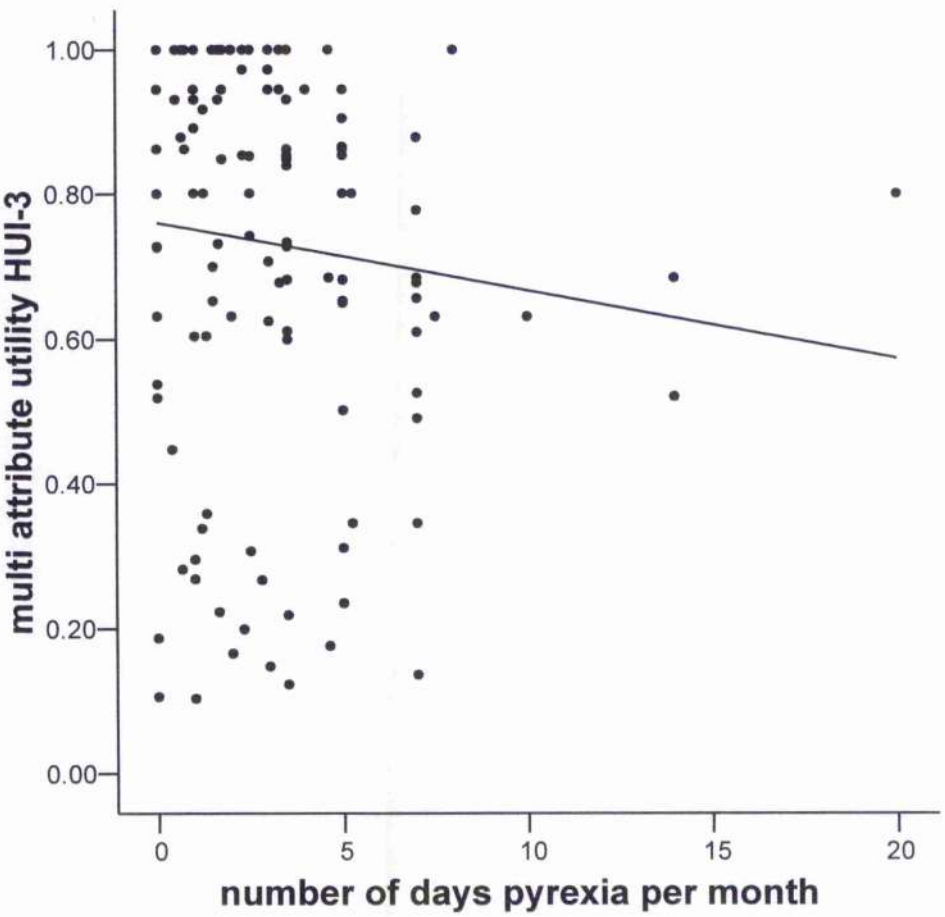


Figure 5.5 HUI multi attribute utility and frequency of sore throat

The relationship between multi-attribute utility and frequency of sore throats in the children with recurrent sore throats (Mann-Whitney, p=0.001).

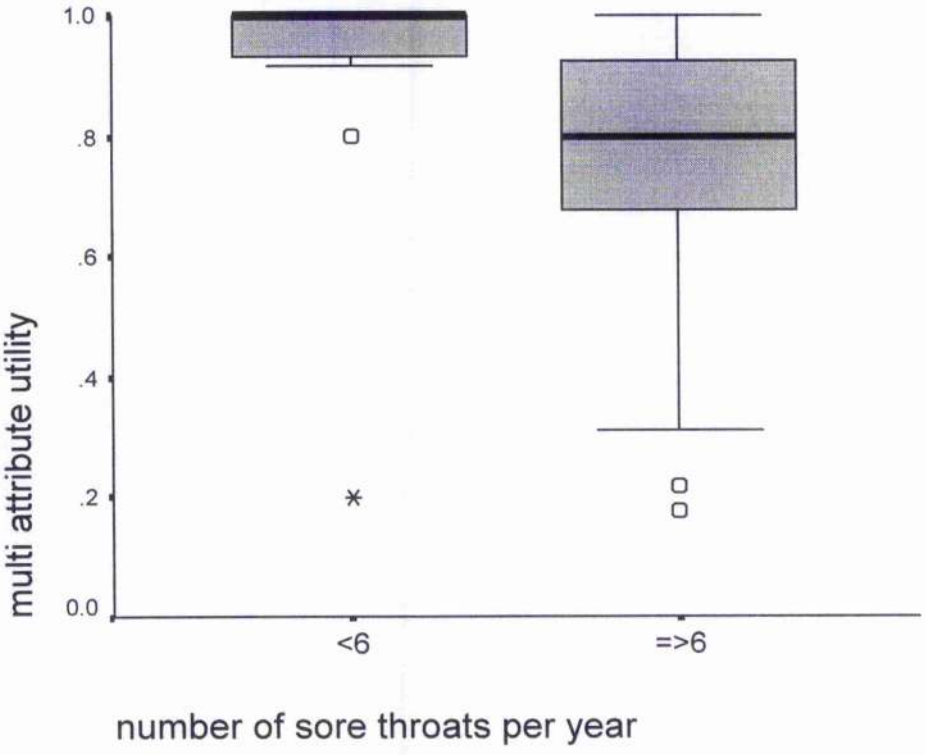
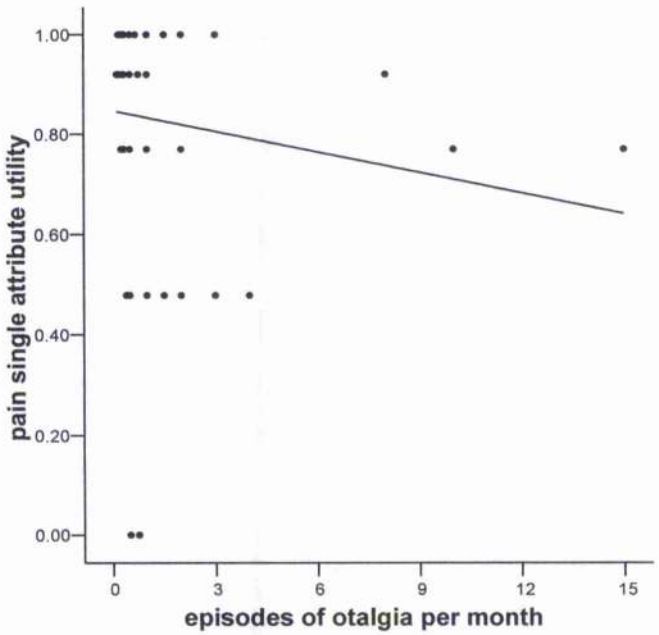


Figure 5.6 HUI pain single attribute utility and frequency of otalgia

There is a weak relationship between the pain single attribute utility and frequency of otalgia in the children with recurrent AOM (Spearman's rho=-0.276.,p=0.009). The relationship with the multi attribute utility is not significant.



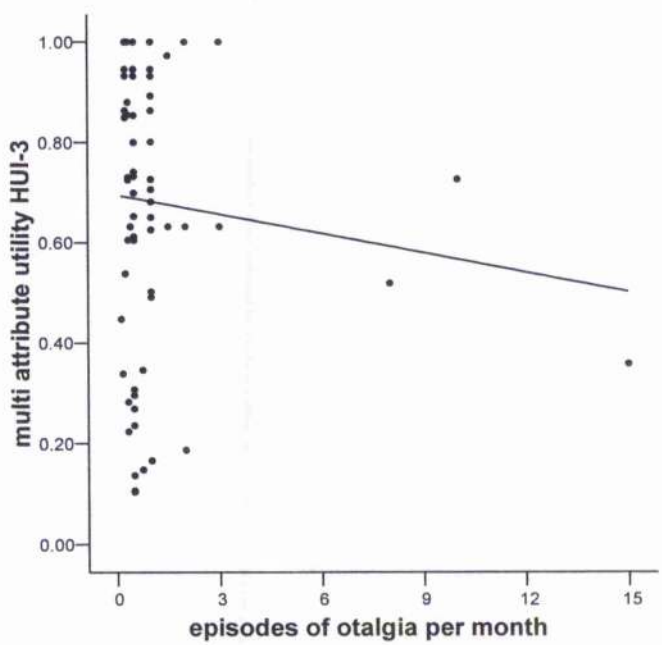


Figure 5.7 HUI hearing single attribute utility and tympanometry

The relationship between presence of bilateral middle ear fluid (type B or C2 tympanograms) and hearing single-attribute utitlity in children referred with suspected OME (Mann-Whitney, p=0.005).

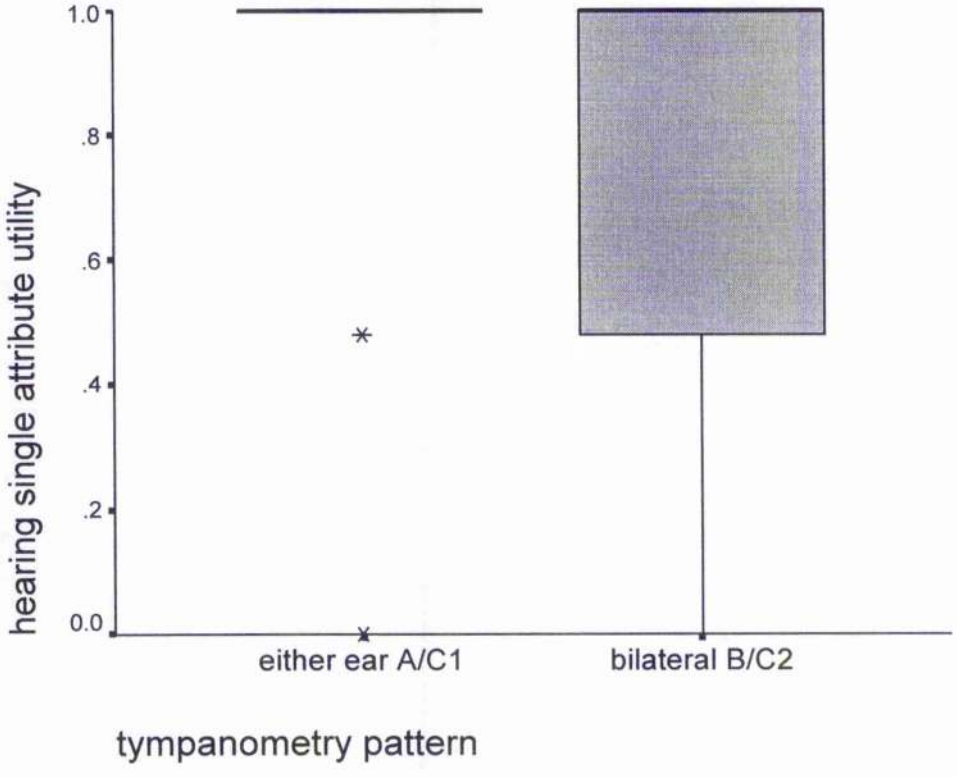
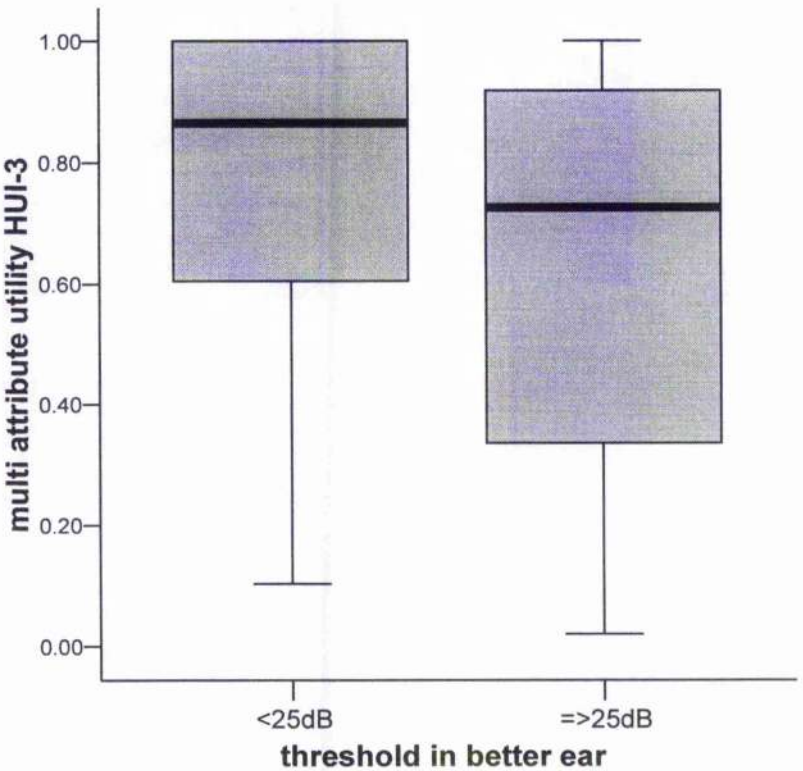
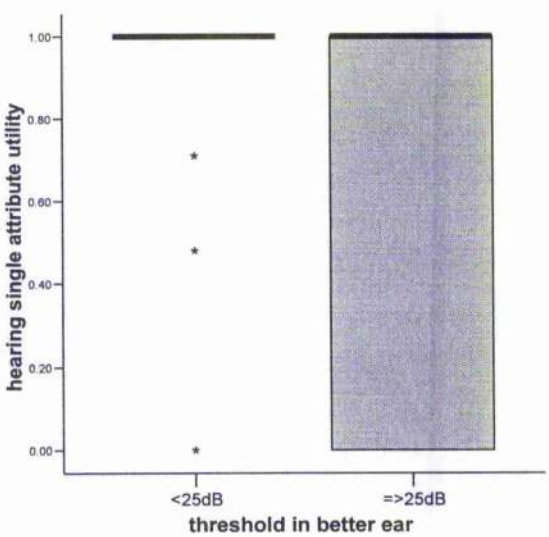


Figure 5.8 HUI utilities and hearing impairment

Boxplots to show that HUI multi attribute utilitiy and single attribute utilities for speech and hearing were worse for those children with OME who had a better-ear threshold of 25dB or worse compared with those who had better hearing (Mann Whitney, p=0.017, 0.003 and 0.025 respectively).





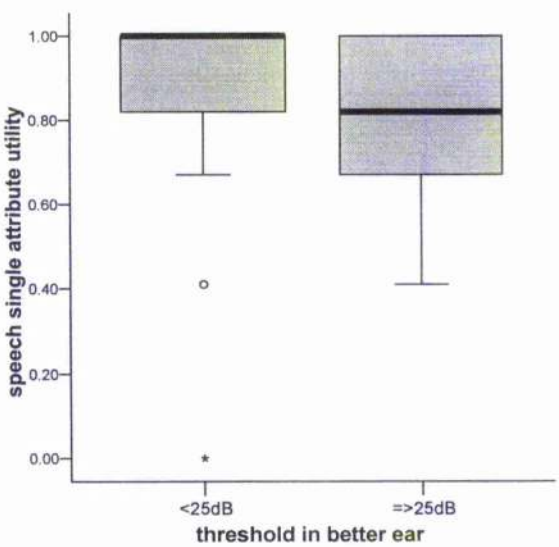


Figure 5.9 HUI multi attribute utility and OM6

The correlation between multi-attribute utility and ear-related handicap, as rated on the OM6 questionnaire (Spearman's rho=-0.608, p<0.001).

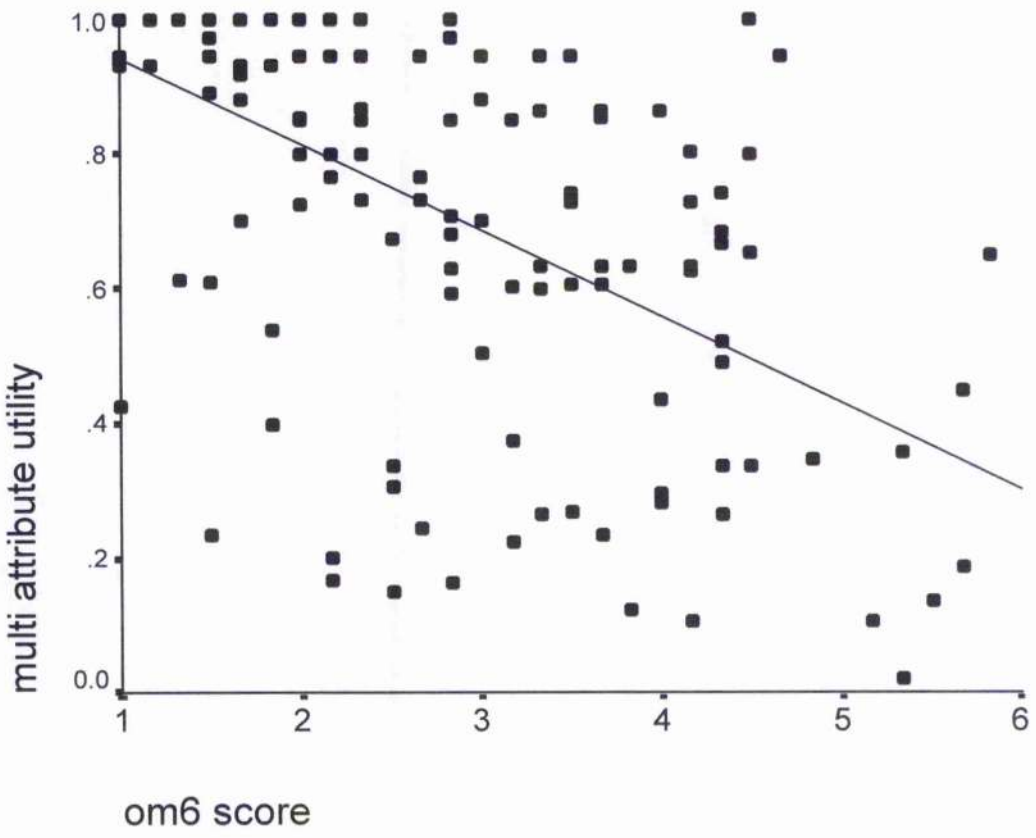


Figure 5.10 HUI multi attribute utility and overall HRQOL (VAS)

The correlation between the HUI multi-attribute utility and overall health-related quality of life, rated on a 100mm visual analogue scale anchored with "totally normal, no problems at all" at 0mm and "worst possible, life totally ruined" at 100mm (Spearman's rho=-0.408, p<0.001).

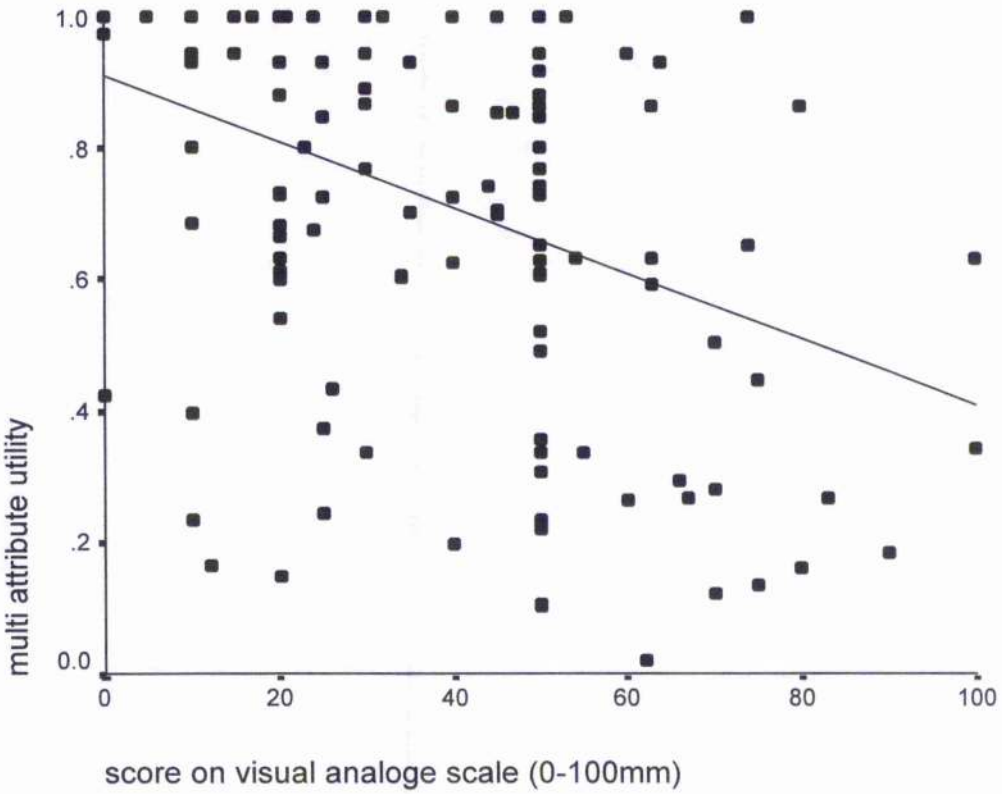


Figure 5.11 HUI multi attribute utility and overall HRQOL (Likert scale)

The association between the HUI multi-attribute utility and overall ratings of health-related quality of life on a five-point scale, with 1 being "excellent" and 5 being "very poor" (Jonckheere-Terpstra, $p<0.001$).

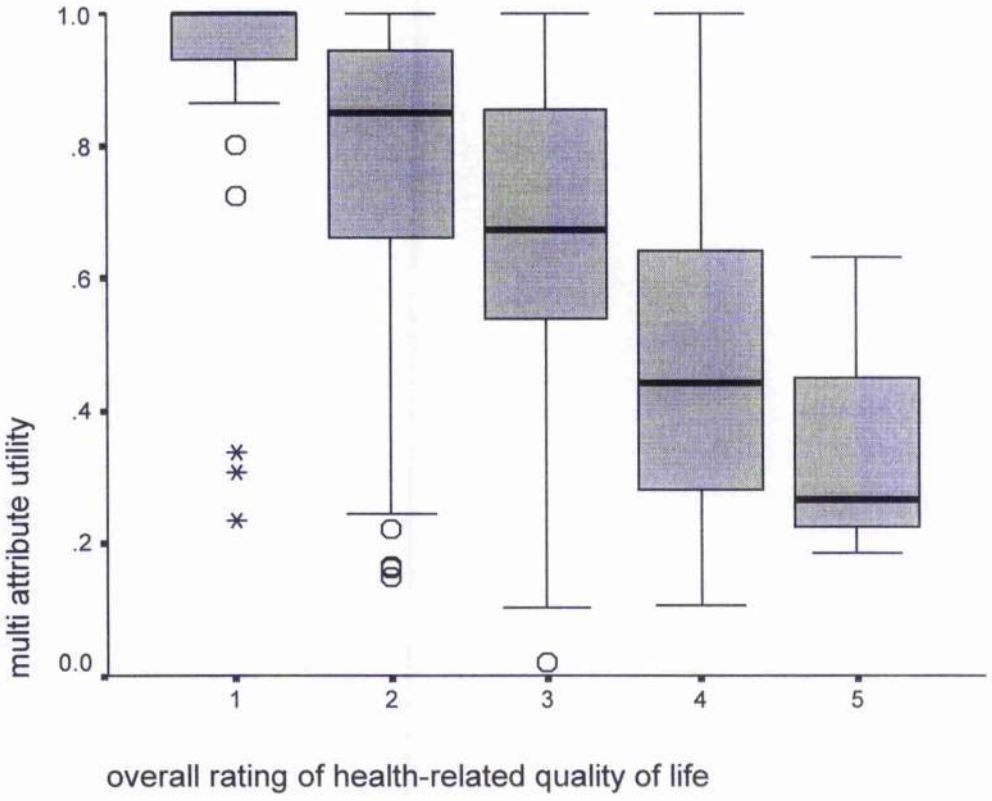


Figure 5.12 Ceiling effects in CHQ domain scores

Examples of four CHQ domains, two of which show significant ceiling effects.

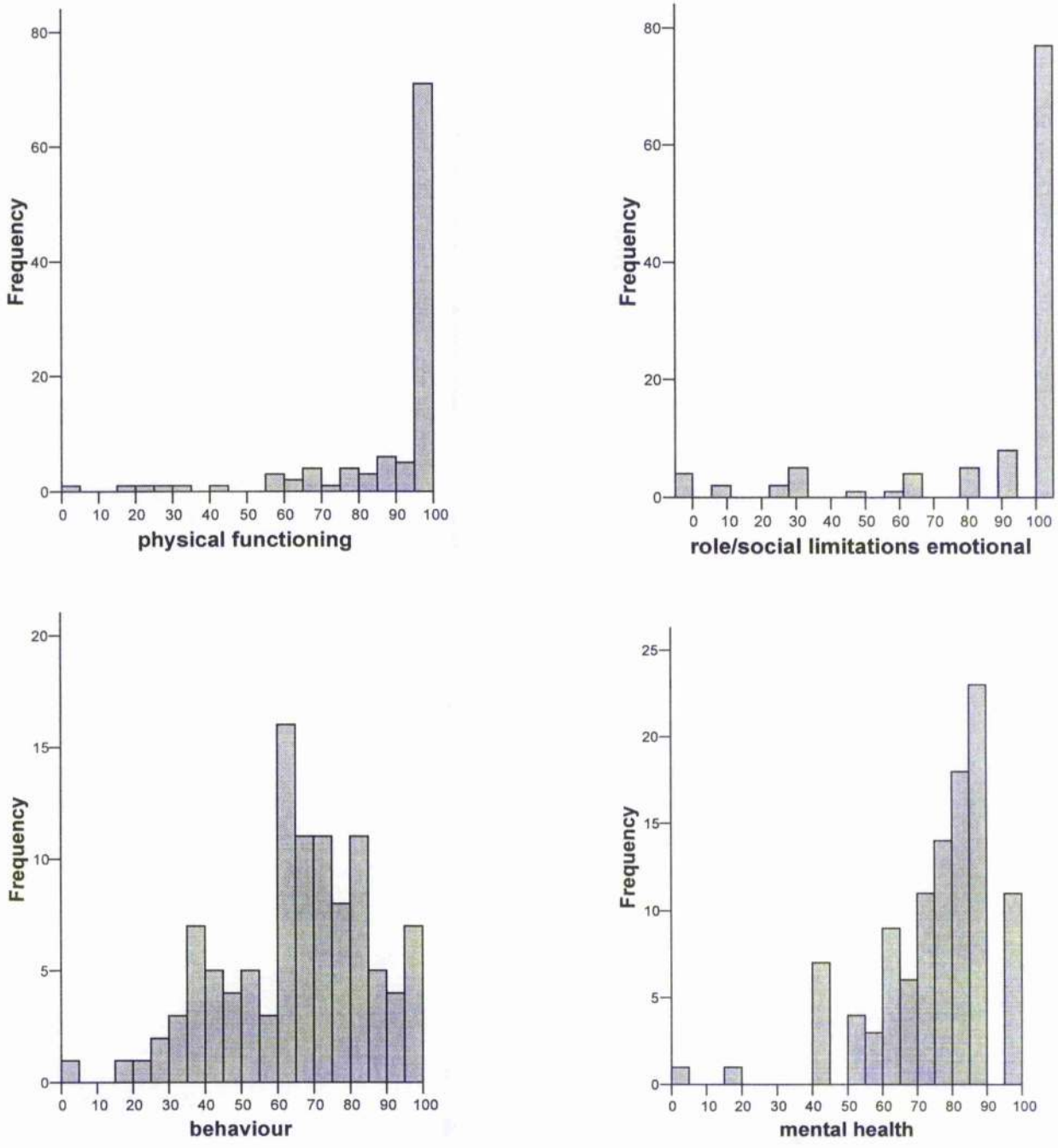


Figure 5.13 CHQ summary / domain scores and referral diagnosis

Boxplots showing both summary scores of the CHQ, together with two of the domain scores for which children with recurrent AOM scored more poorly than children with OME or sore throats.

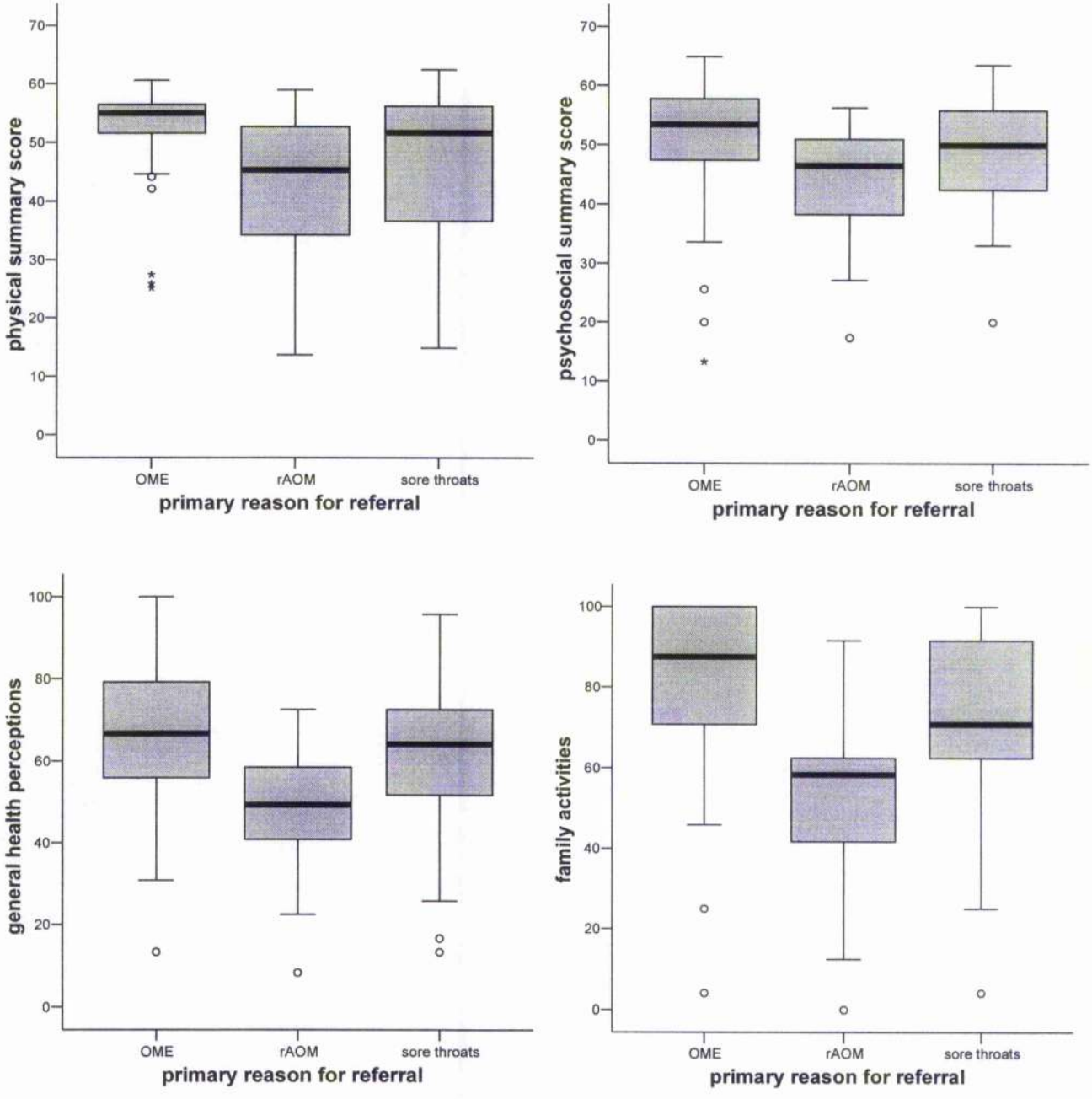
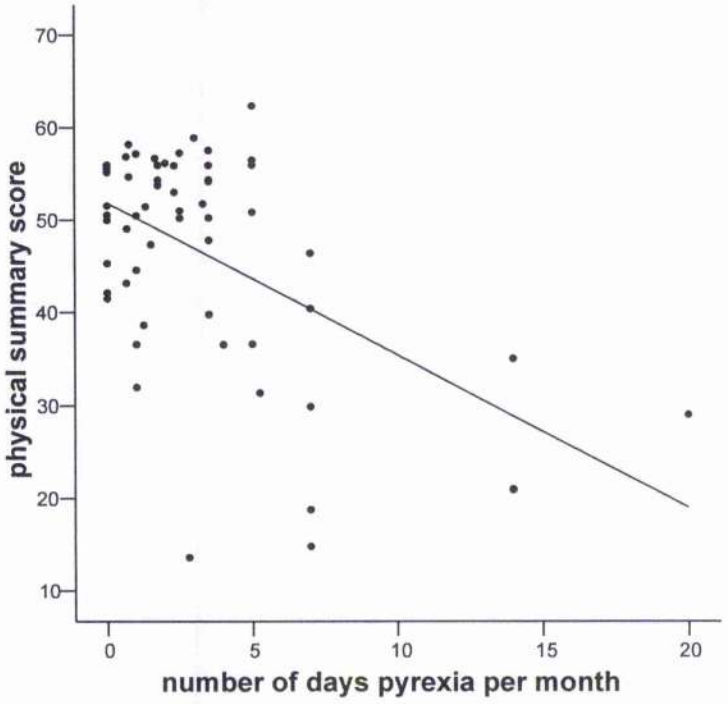


Figure 5.14 CHQ summary scores and frequency of pyrexia

Scatterplots showing the relationship between the CHQ summary scores and the average number of pyrexial illnesses per month in children with recurrent AOM or sore throats (Spearman's rho= -0.311 and -0.394, p=0.018 and 0.002 respectively).



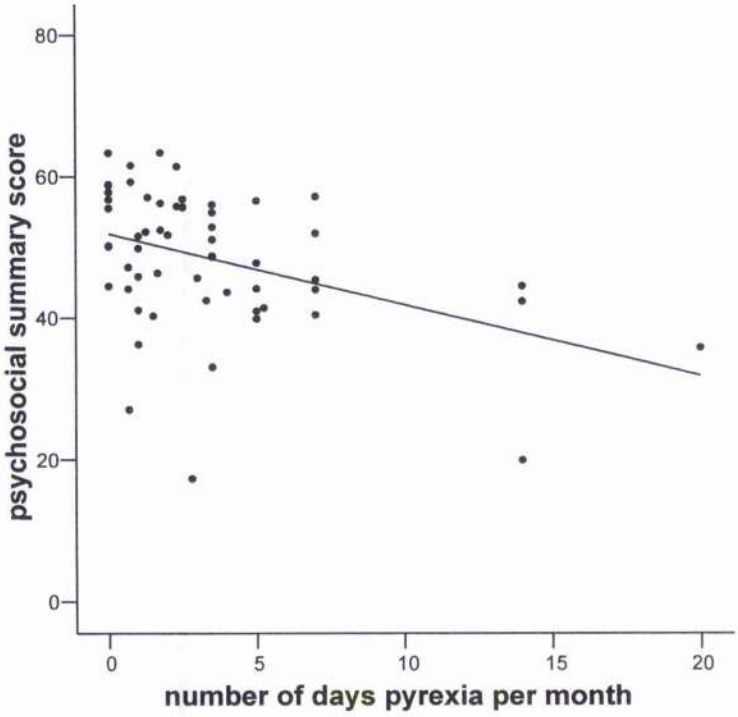
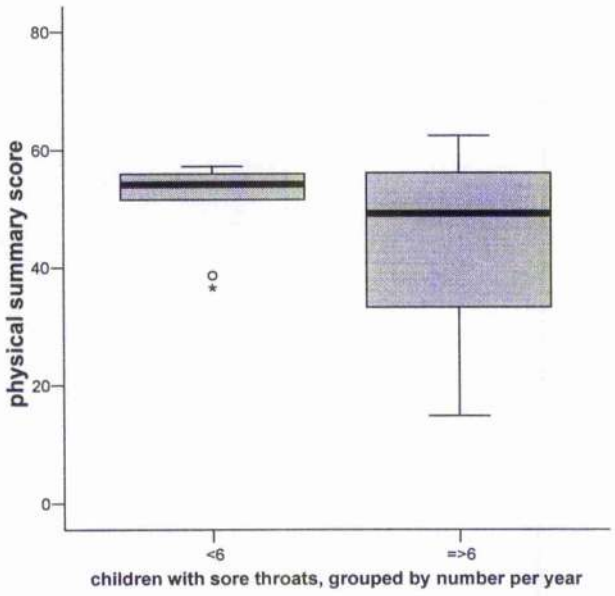
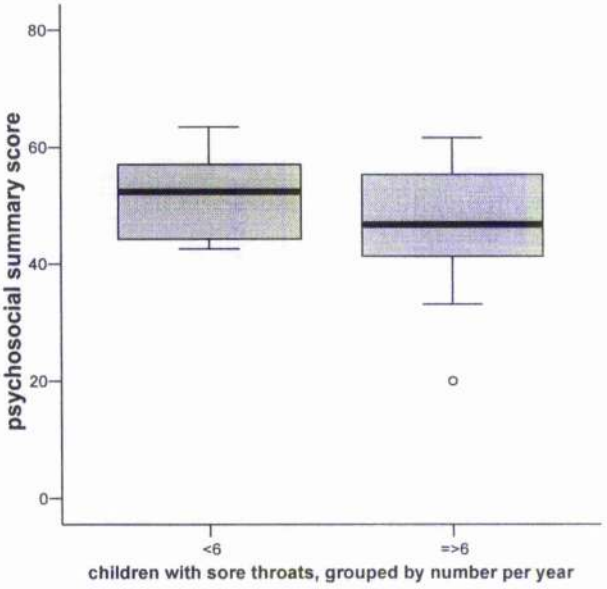
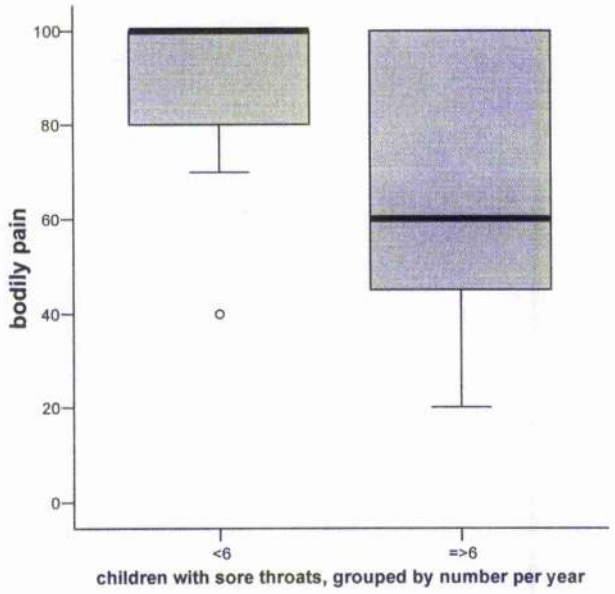


Figure 5.15 CHQ summary / domain scores and frequency of sore throat

Boxplots showing the relationship between the summary scores and two of the domain scores of the CHQ and frequency of sore throats (median 6 per year).







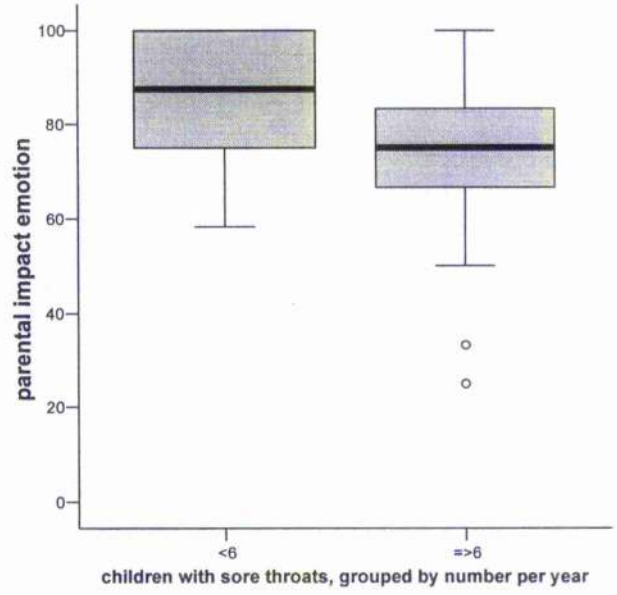
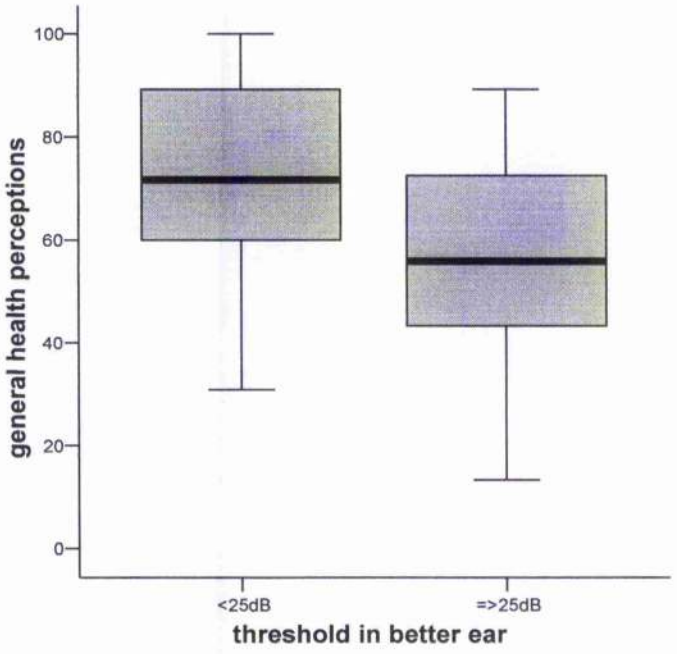


Figure 5.16 CHQ General Health Perceptions: hearing and tympanometry

The relationship between the General Health Perceptions domain of the CHQ and markers of disease severity in OME: presence of a better ear threshold of 25dB or worse (Mann Whitney, p=0.008) and presence of bilateral B/C2 tympanometry (p=0.013).



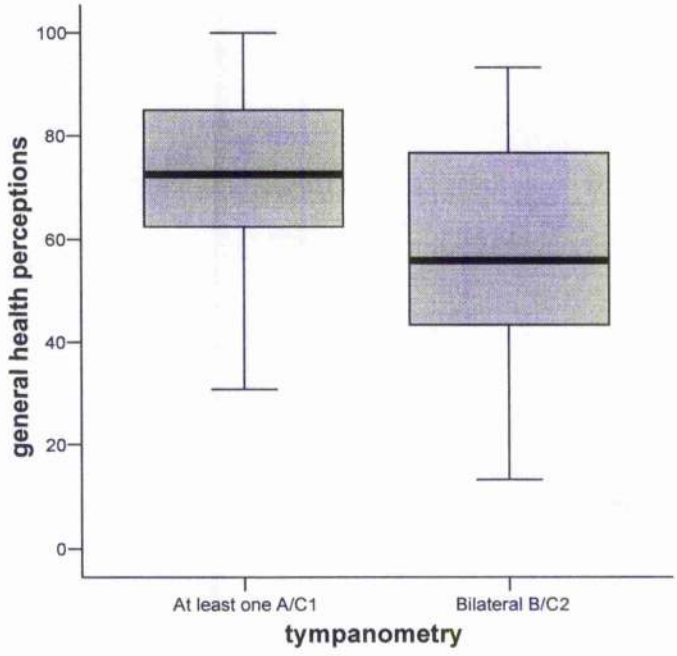


Figure 5.17 CHQ summary scores and OM6

Scatterplots showing the relationship between the summary scores of the CHQ and ear-related handicap rated using OM6 in children with OME or recurrent AOM (Spearman's rho=-0.373 and -0.404, p=0.002 and 0.001 respectively).

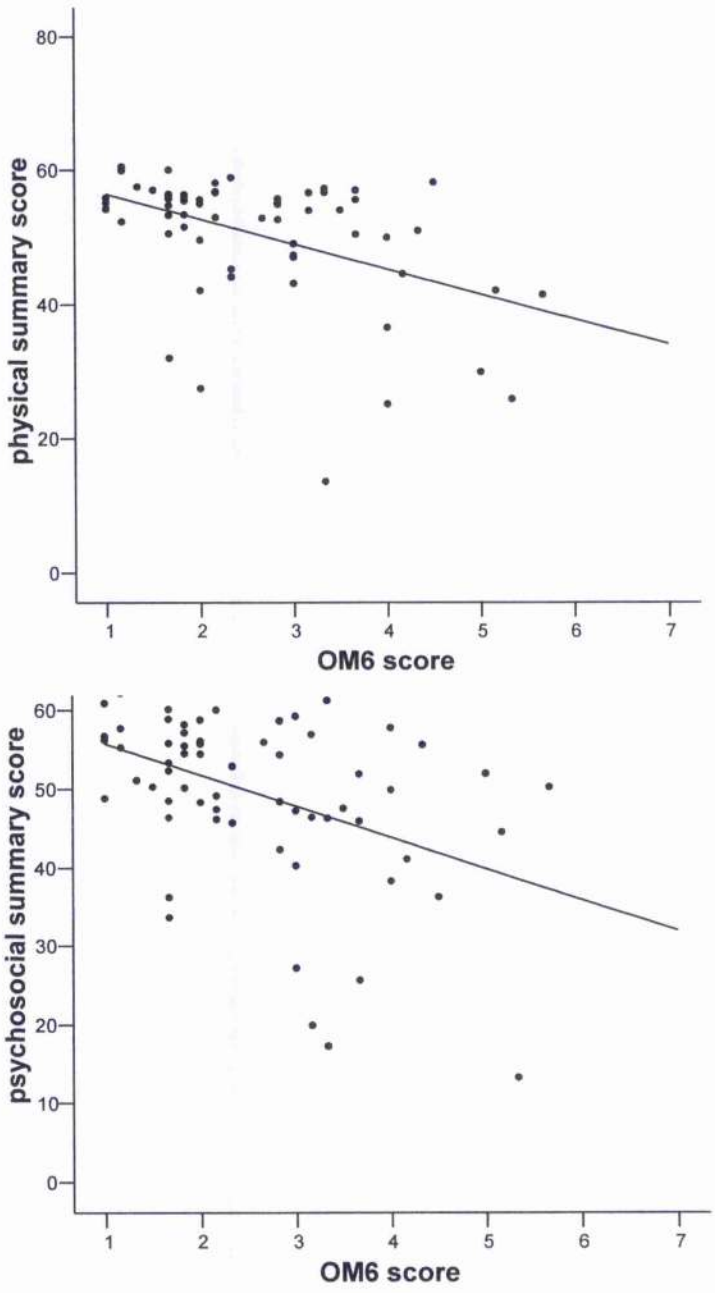


Figure 5.18 CHQ summary scores and overall HRQOL (visual analogue)

The relationship between the CHQ summary scores and overall HRQOL directly rated on a 100mm visual analogue scale (Spearman's rho=-0.410 and -0.226, p= <0.001 and 0.026 respectively).

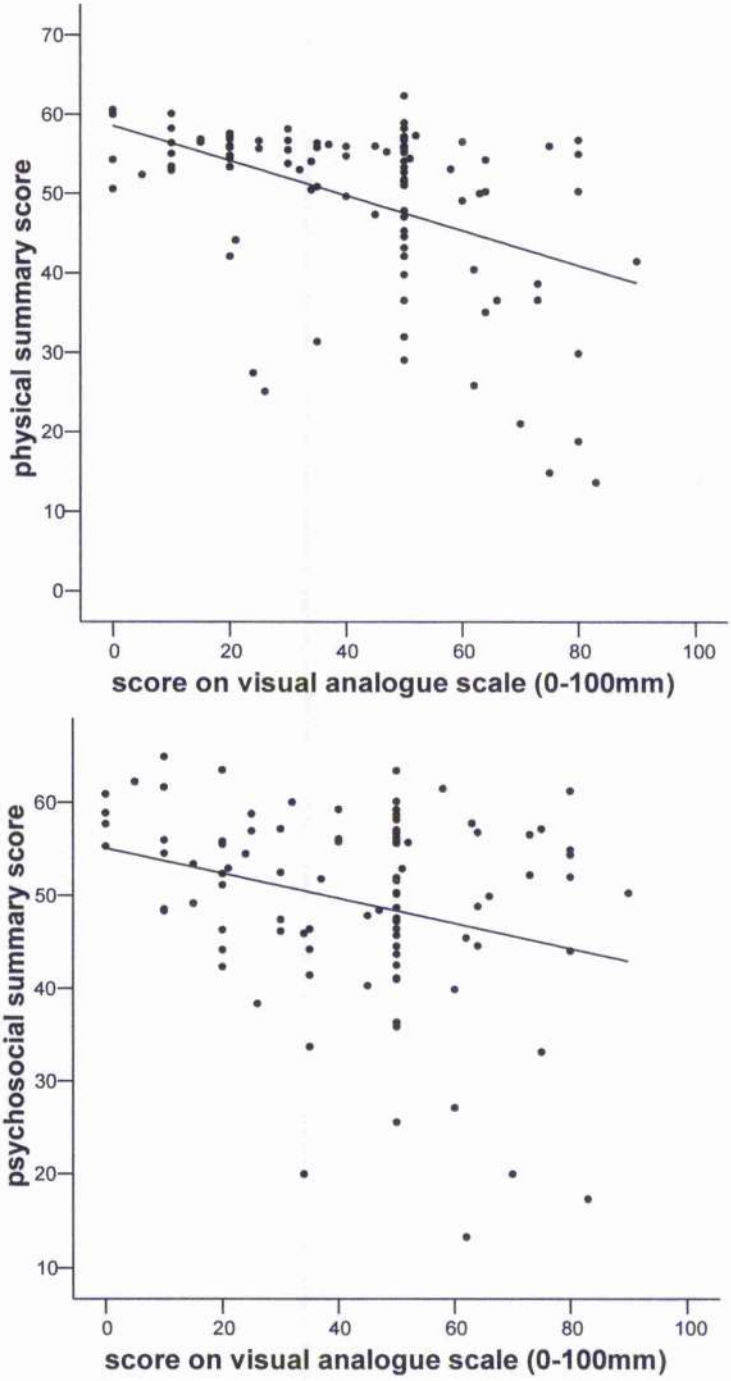


Figure 5.19 CHQ summary scores and overall HRQOL (Likert scale)

The relationship between CHQ summary scores and overall HRQOL directly rated on a 5-point scale, with 1 being "excellent" and 5 being "very poor" (Jonckheere-Terpstra, $p<0.001$ in both cases).

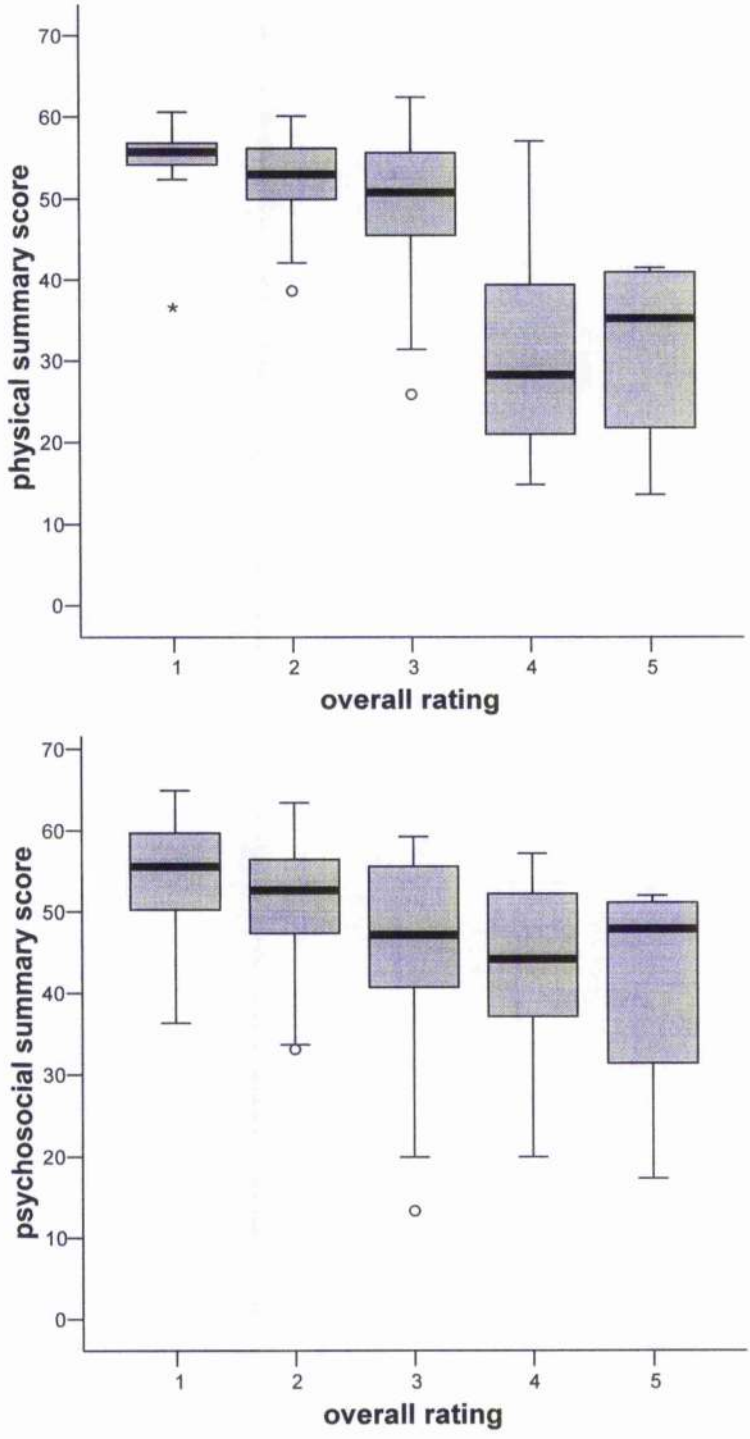


Figure 5.20 Ceiling effect in TACQOL Cognition domain score

The distribution of scores in the TACQOL Cognition domain is shown as a histogram as an example of a domain with a ceiling effect in the population studied. The Negative Emotions domain shows no ceiling effect.

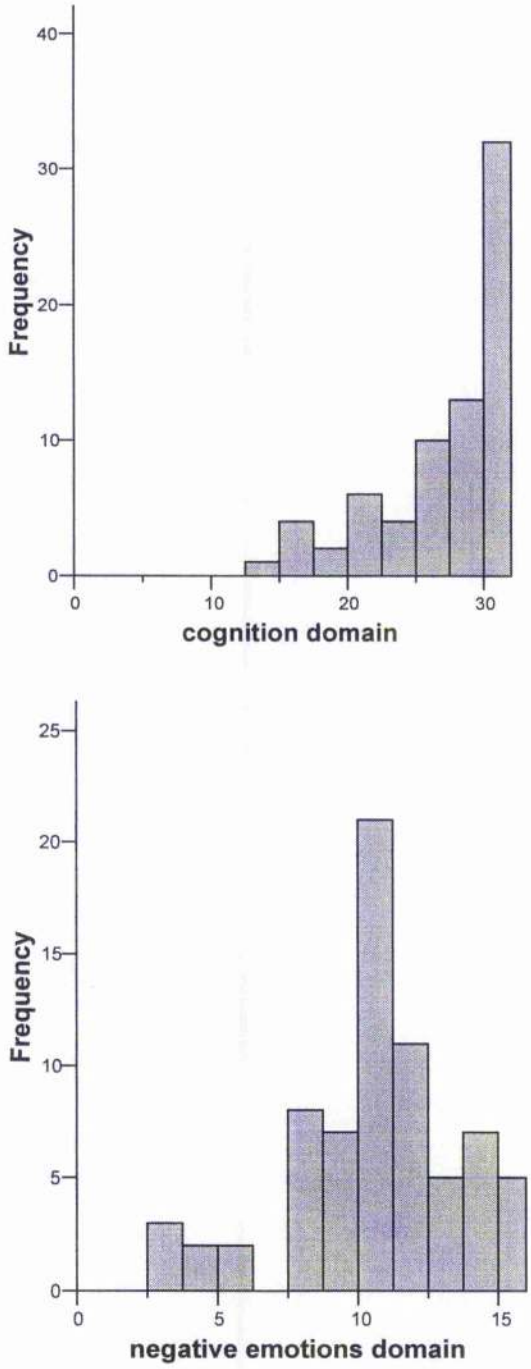
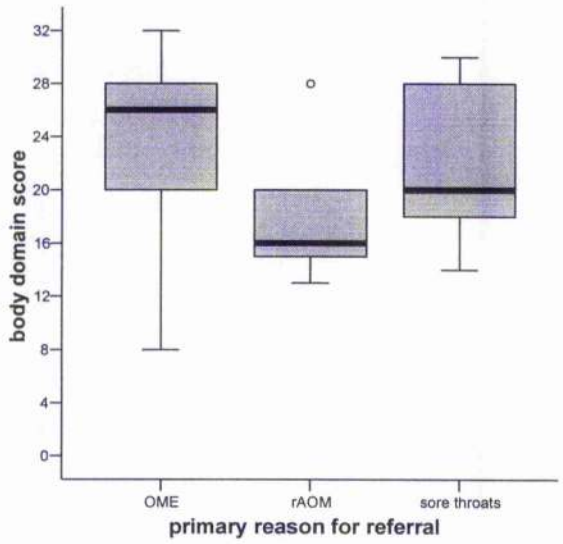
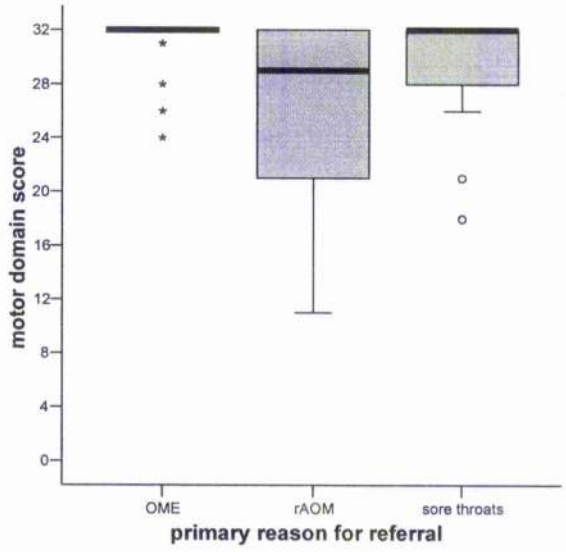


Figure 5.21 TACQOL domain scores and referral diagnosis

Boxplots showing that the children with recurrent AOM have worse domain scores for Body, Motor and Autonomy than those with either OME or sore throats (Kruskal Wallis, p=0.015, 0.02 and 0.006 respectively).





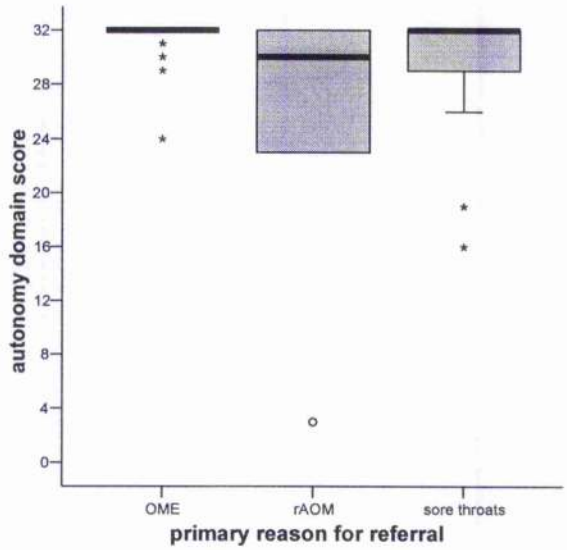
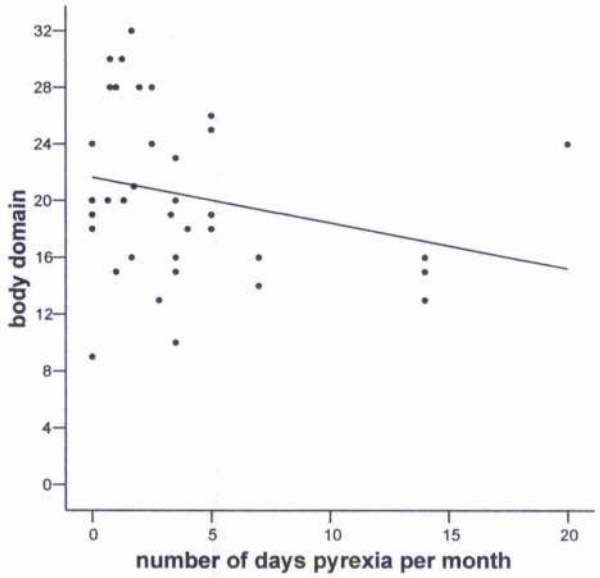


Figure 5.22 TACQOL domain scores and frequency of pyrexia

Scatterplots of the average number of days per month with pyrexial illness in children with recurrent AOM or sore throats against the TACQOL domain scores Body and Positive Emotions (Spearman's rho=-0.327 and -0.340, p=0.045 and 0.034 respectively).



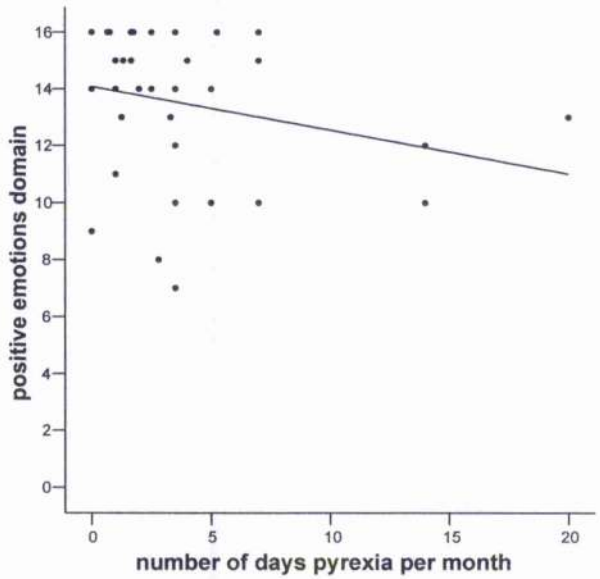


Figure 5.23 TACQOL body domain score and frequency of sore throat

Scatterplot of frequency of sore throat against TACQOL body domain score. The number of children studied was small, and the correlation is not statistically significant (n=27, Spearman's rho=-0.369, p=0.058).

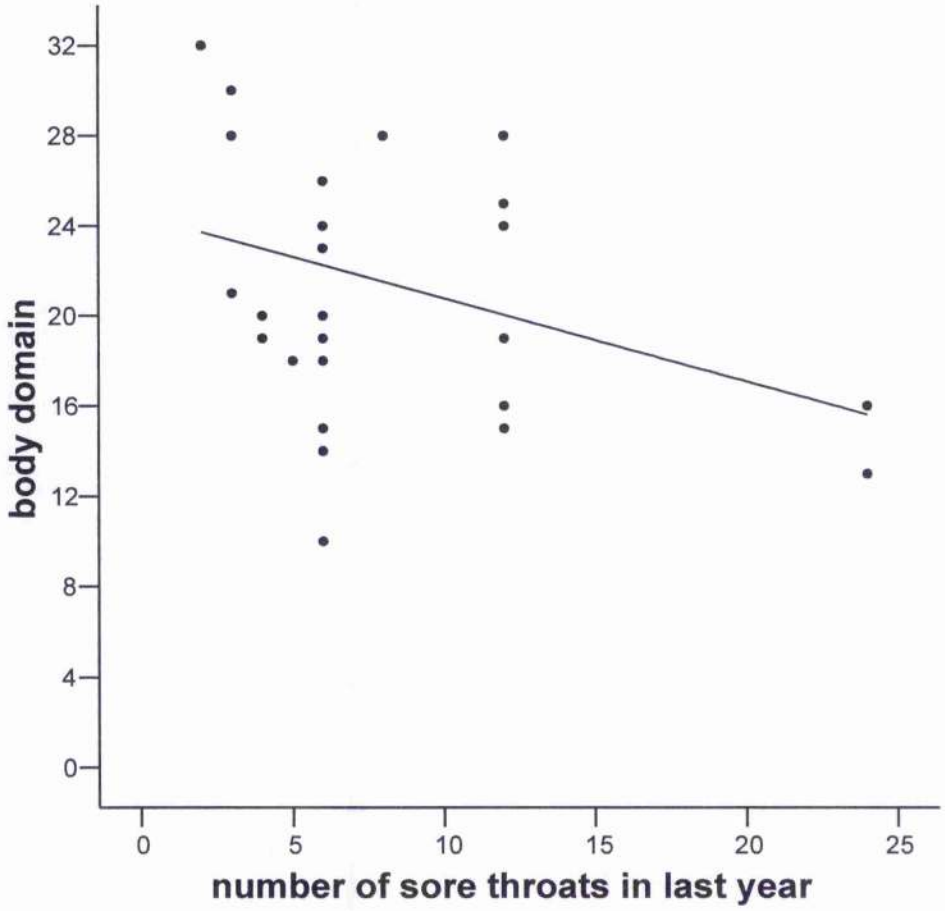


Figure 5.24 TACQOL body domain score and tympanometry

Boxplot showing that, for children with OME, the Body domain score was associated with the presence of type B/C2 tympanometry (Mann Whitney, p=0.035).

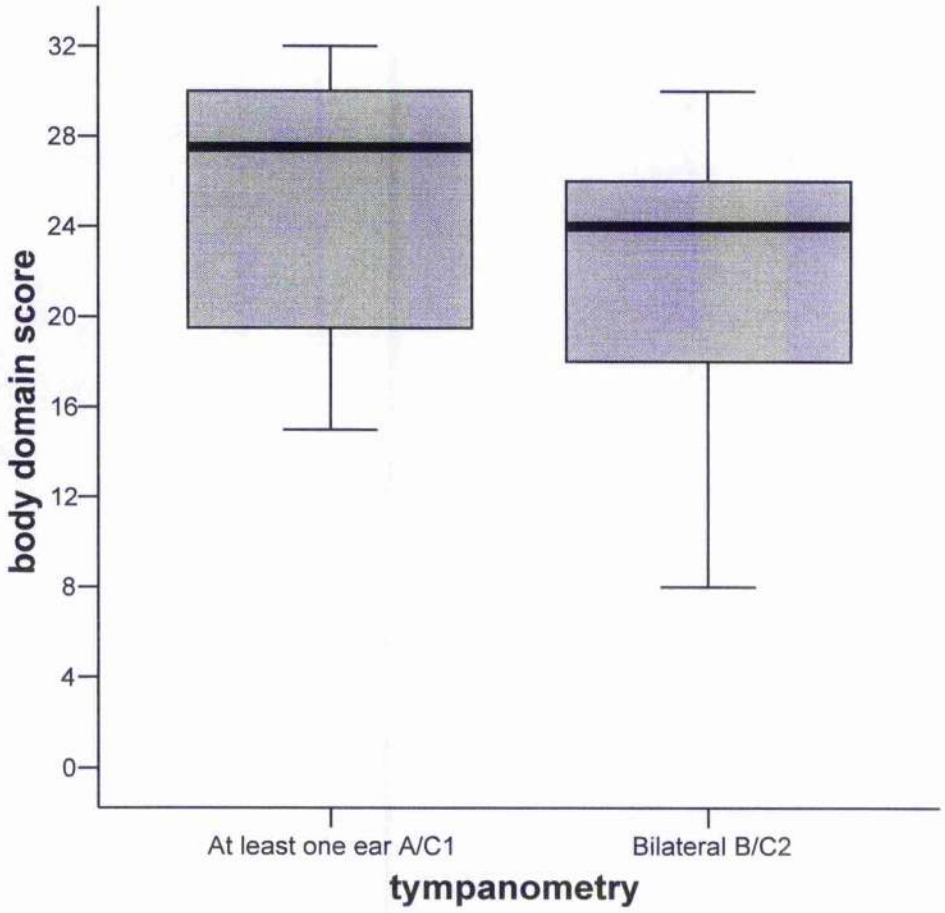


Figure 5.25 TACQOL domain scores and OM6

Scatterplots showing the correlation between ear-related handicap rated using OM6 and two of the TACQOL domain scores, Body and Cognition (Spearman's rho=-0.569 and -0.401, p= <0.001 and 0.006 respectively) in children with OME or recurrent AOM.

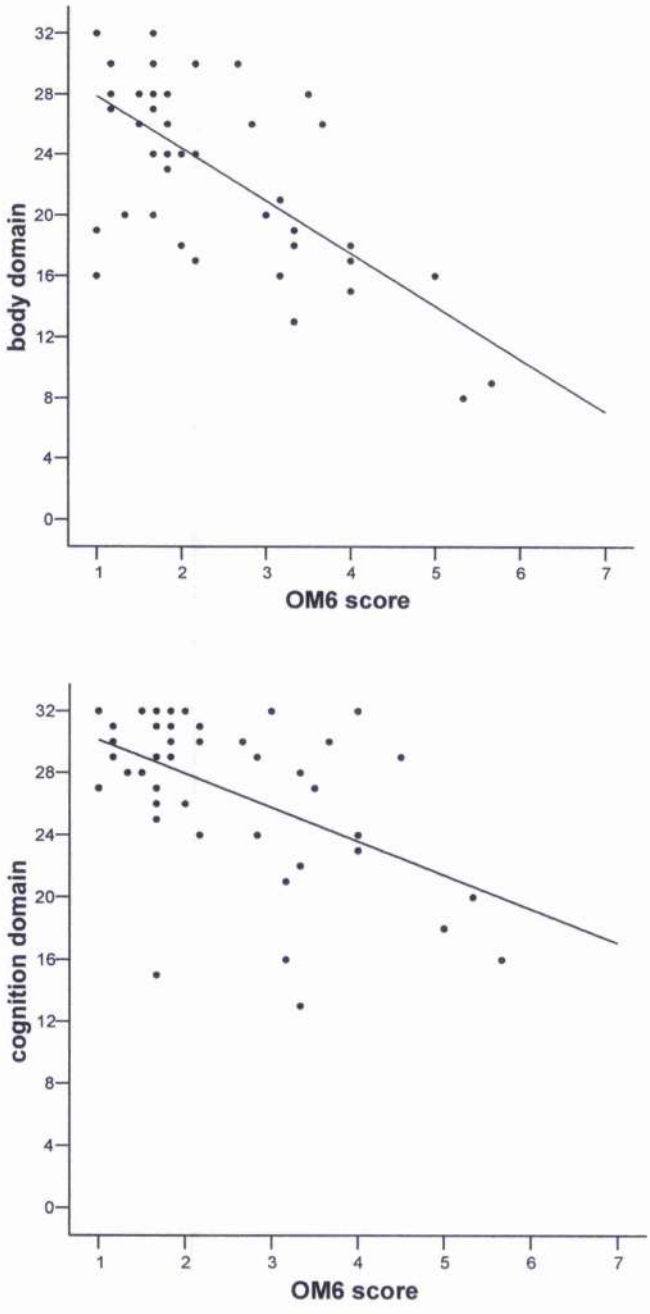


Figure 5.26 TACQOL domain scores and overall HRQOL (Likert scale)

The association between four of the TACQOL domain scores and overall ratings of health-related quality of life on a five-point scale, with 1 being "excellent" and 5 being "very poor" (see Table 7.15 for Jonckheere-Terpstra test of statistical significance).

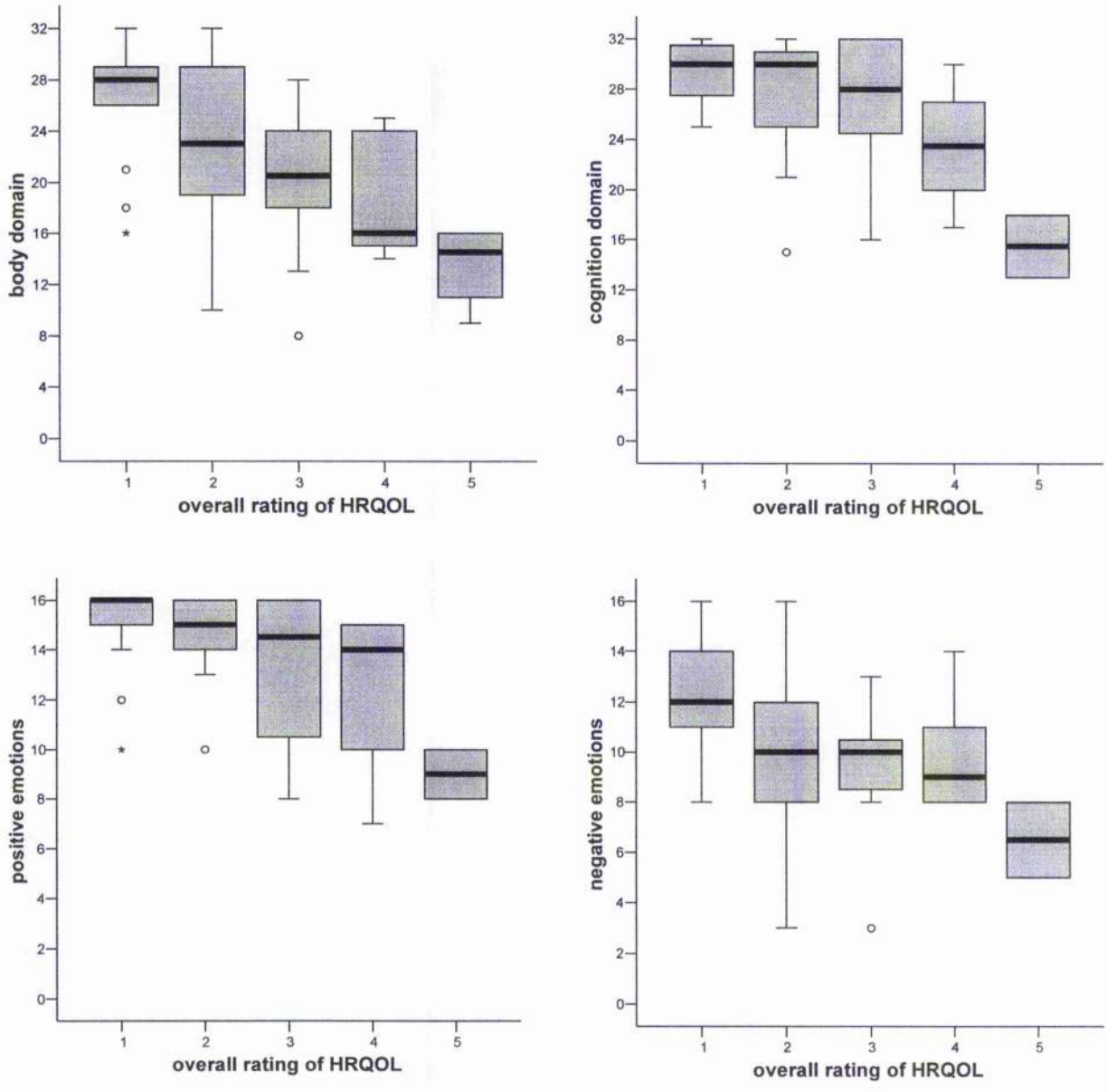


Figure 5.27 Ceiling effect in TAPQOL skin domain score

The distribution of scores in the TAPQOL Skin domain is shown as a histogram as an example of a domain with a ceiling effect in the population studied.

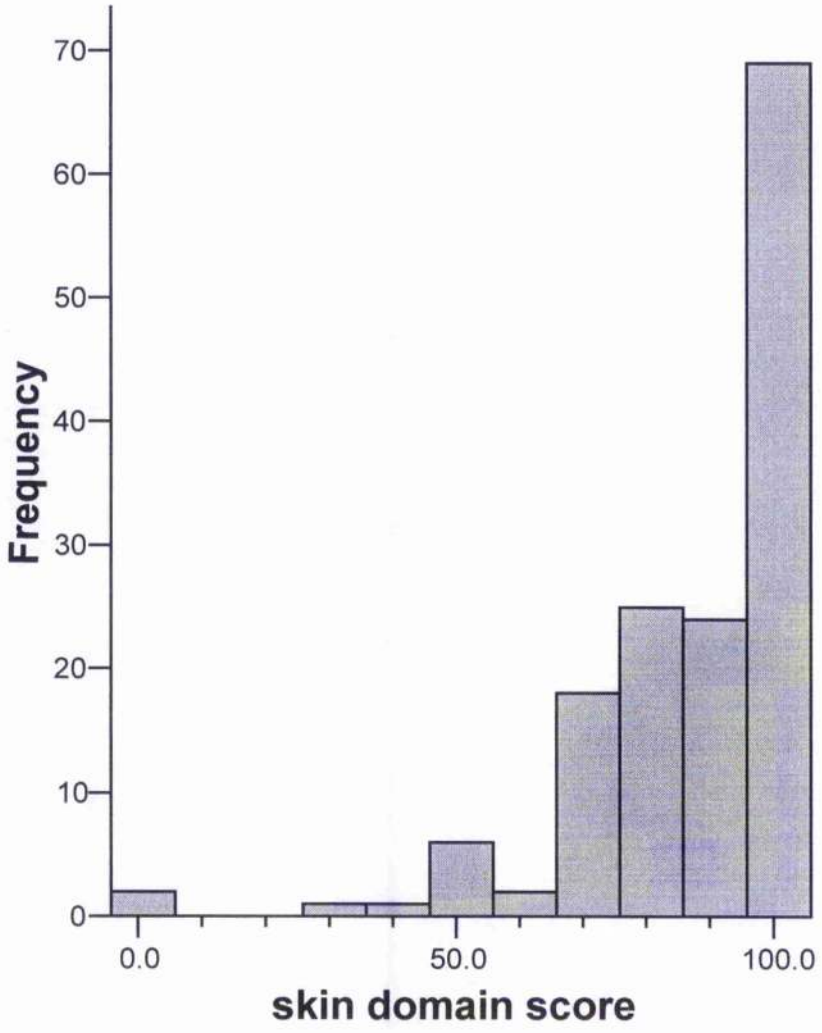
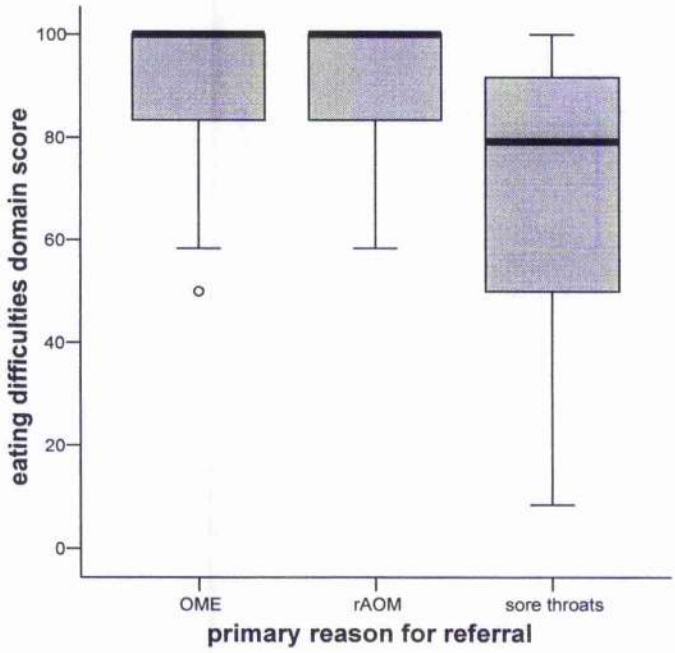


Figure 5.28 TAPQOL domain scores and referral diagnosis

Boxpots showing the Appetite and Eating Difficulties domain scores, both of which were significantly worse in children with sore throats (Kruskal Wallis, $p<0.001$).



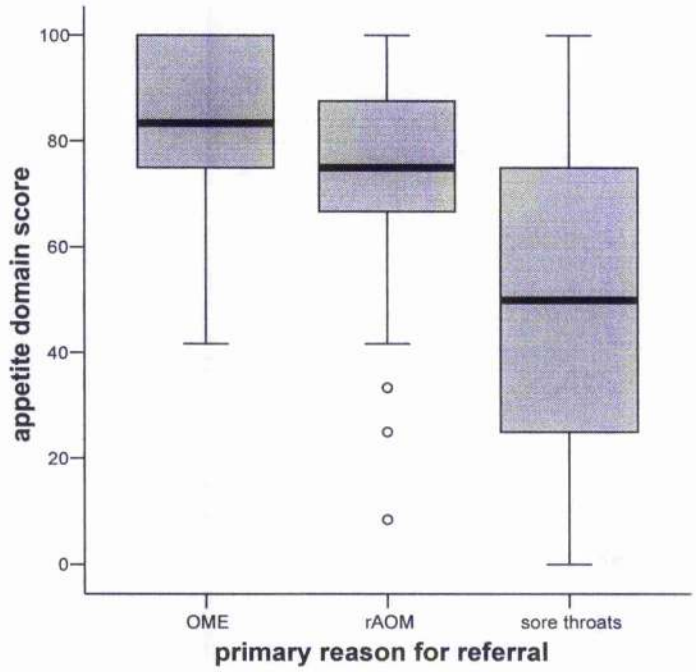


Figure 5.29 TAPQOL Appetite domain score and frequency of pyrexia

An example of the degree to which TAPQOL domain scores (in this case, appetite) correlate with the frequency of pyrexial illness in children with recurrent AOM or sore throats (Spearman's rho=-0.465, p<0.001).

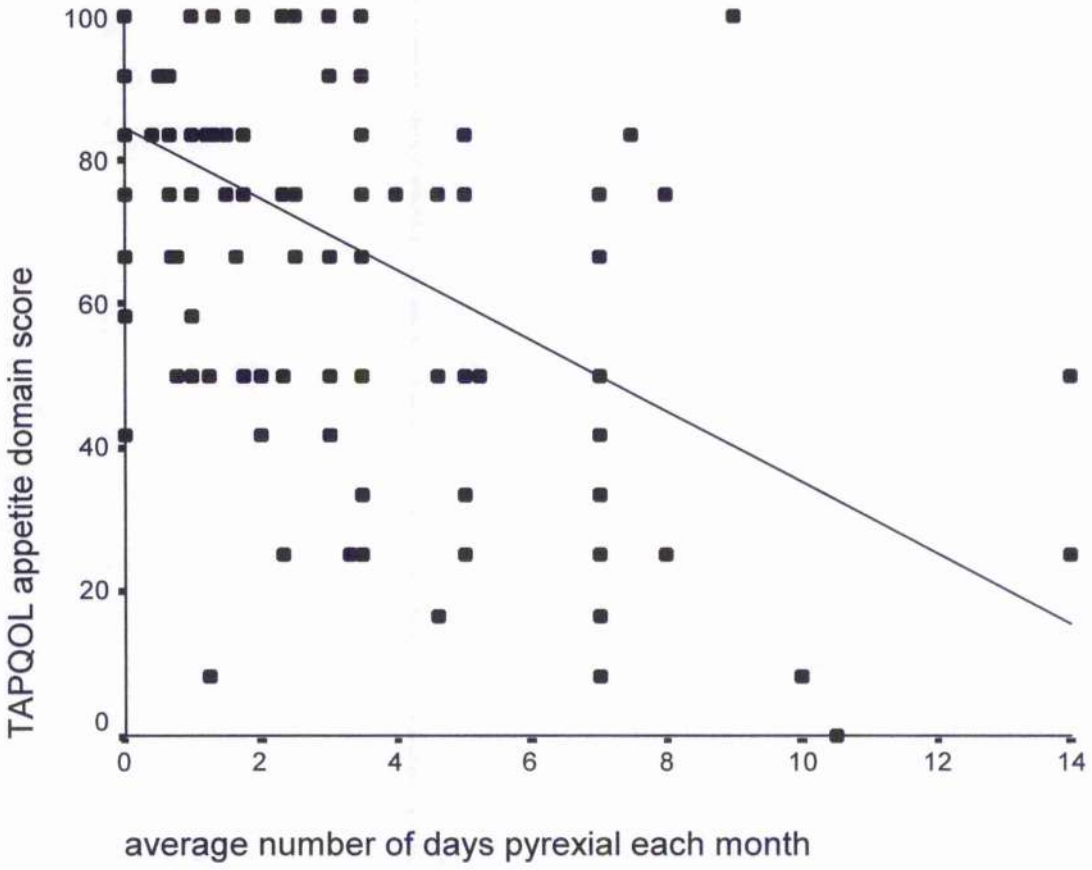


Figure 5.30 TAPQOL Communication domain score and tympanometry

TAPQOL domain scores in children with OME were not any worse in those children who had bilateral B or C2 tympanograms (confirming the presence of middle ear effusion), compared with those children who had tympanometry showing at least one ear to be clear of fluid (Mann-Whitney, p=0.532).

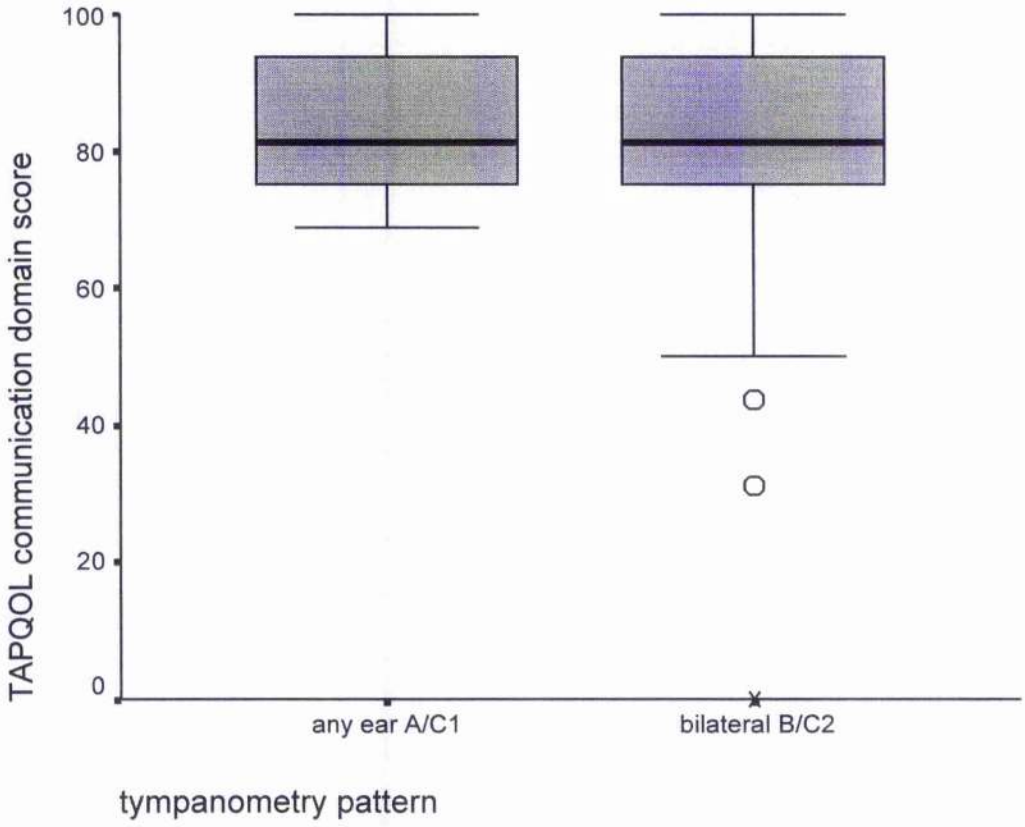


Figure 5.31 TAPQOL Sleep domain score and OM6

There is some correlation between TAPQOL domain scores and ear-related handicap, as measured with the OM6 questionnaire (Spearman's rho=-0.339, $p<0.001$).

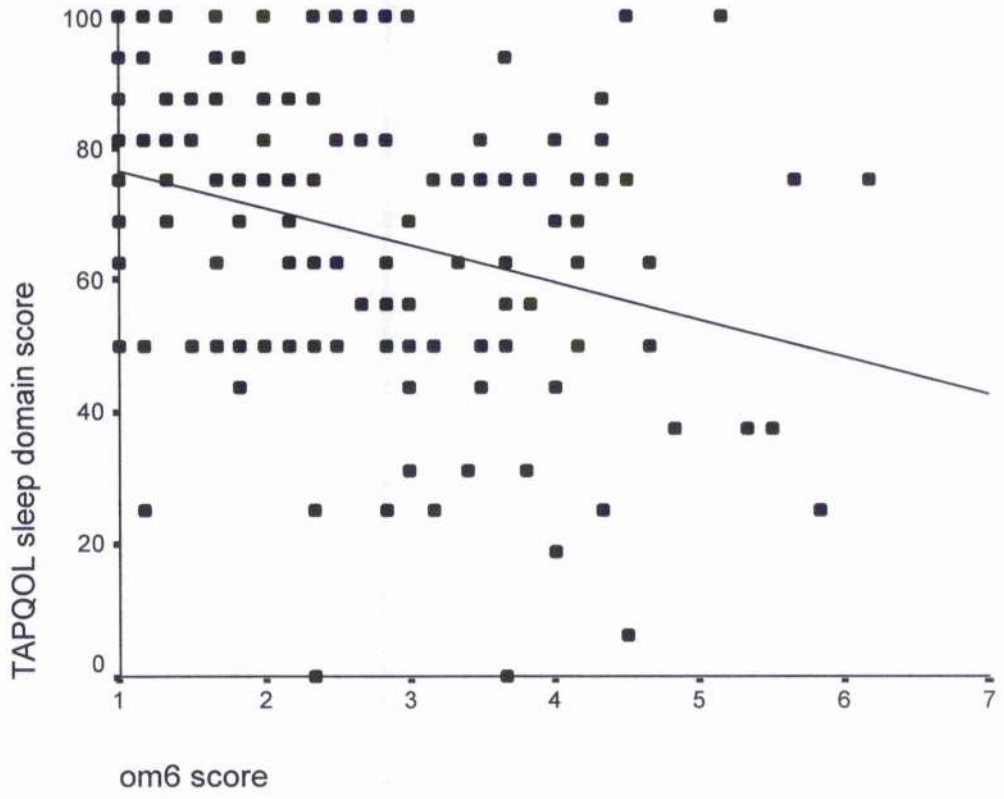


Figure 5.32 TAPQOL Appetite domain score and overall HRQOL

TAPQOL domain scores were associated with assessments of the child's overall health-related quality of life on a 5-point Likert rating scale, with 5 representing "poor" and 1 "excellent" quality of life (Jonckheere-Terpstra test, $p<0.001$).

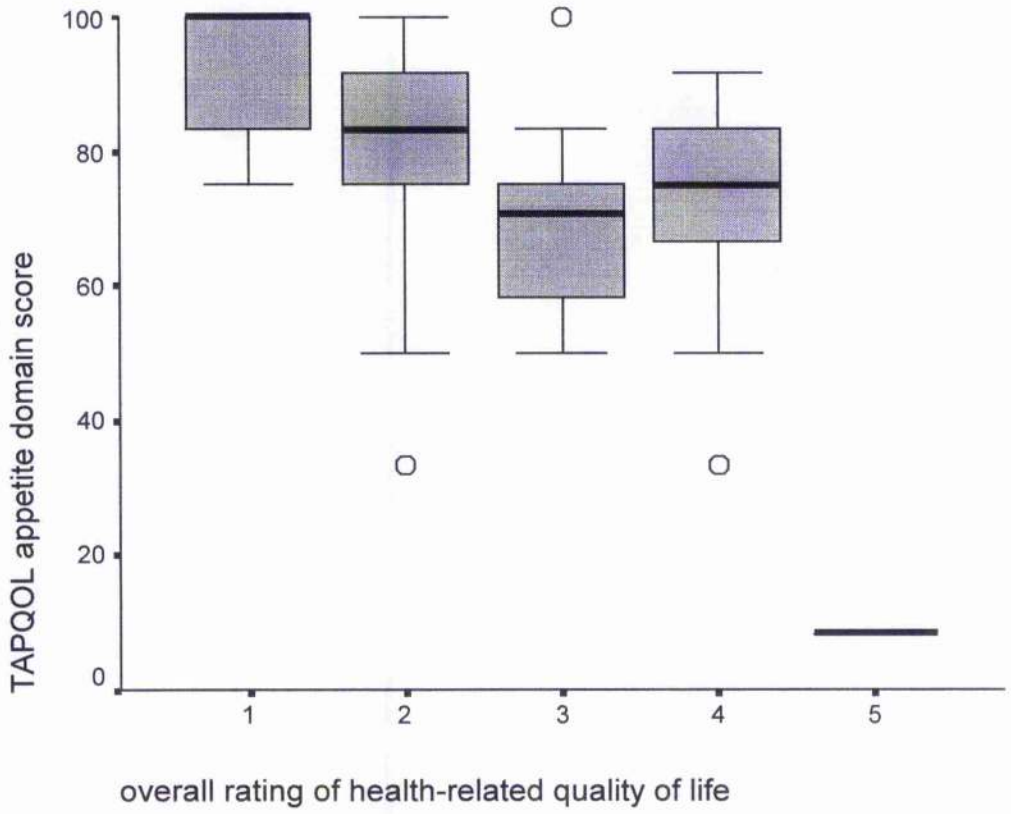


Figure 5.33 TAPQOL Communication domain score and HUI mark III

TAPQOL domain scores were correlated with the child's overall health-related quality of life assessed using the Health Utilities Index mark III questionnaire (Spearman's rho=0.582, p<0.001).

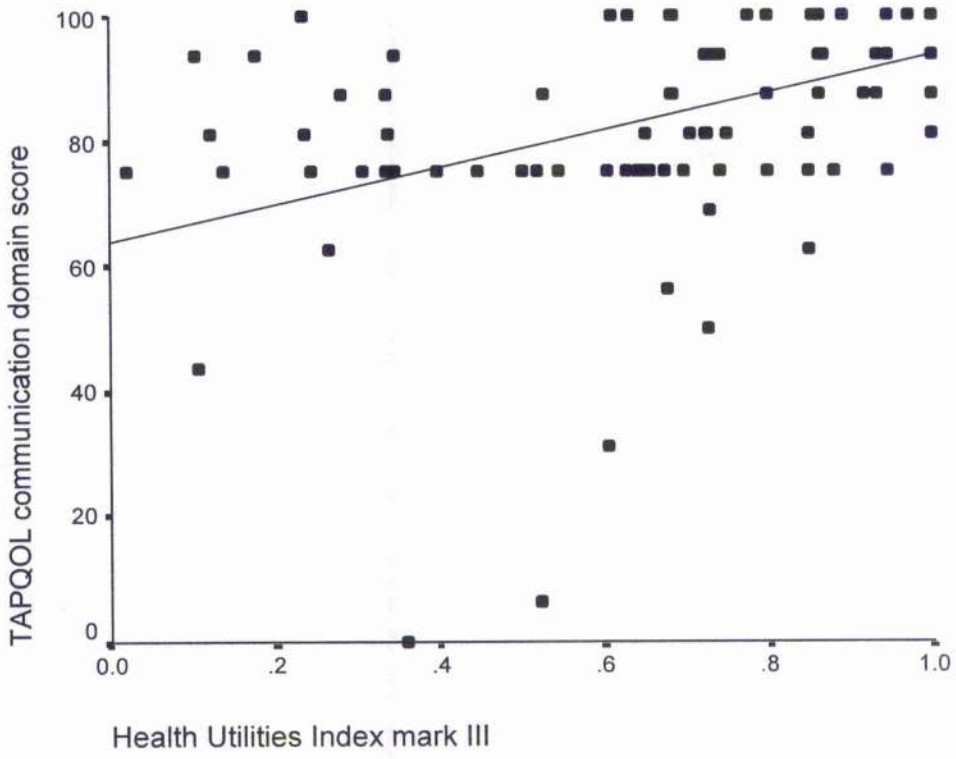


Table 5.1 HUI mark III utility scores

The means, standard deviations (s.d.) and ranges of values obtained for each of the single attribute utilities and the multi attribute utility of the HUI. These values are for the whole group of children in the study (n=252).

Utilities	Min	Mean	Max	s.d.
vision	0.59	0.99	1	0.04
Hearing	0	0.87	1	0.30
speech	0	0.89	1	0.20
Emotion	0.33	0.95	1	0.11
pain	0	0.87	1	0.21
Ambulation	0	0.97	1	0.16
Dexterity	0.20	0.99	1	0.07
Cognition	0	0.93	1	0.18
multi attribute utility	**0.02**	**0.75**	**1**	**0.27**

Table 5.2 HUI utilities: age, sex and deprivation

The effect of age, sex and socio-economic deprivation on HUI multi and single attrbute utilities (*Jonckheere-Terpstra test across 14 groups representing age in years or 5 groups representing quintiles of Carstairs Deprivation Index distribution; **Mann-Whitney test; n=252)

Utilities	Age of child *	Sex of child **	Sex of respondent **	Carstairs Deprivation Index *	Manual vs Non-manual occupation **
vision	0.890	0.803	0.414	**0.012**	**0.024**
Hearing	0.857	0.757	0.427	0.392	0.147
speech	**<0.001**	0.216	0.517	0.655	0.946
Emotion	0.906	0.150	0.736	0.336	**0.003**
pain	0.829	0.320	0.577	0.890	0.280
Ambulation	**<0.001**	0.812	0.219	0.077	**0.036**
Dexterity	0.208	0.594	0.426	0.419	0.853
Cognition	0.835	0.706	**0.042**	0.437	0.613
multi	**0.033**	0.722	0.357	0.475	0.264

Table 5.3 CHQ factor loadings

The table shows the extent to which each of ten domain scores load onto the two factors extracted. Data from the original CHQ validation studies in the USA are shown for comparison [56]. Eigenvalues greater than 0.4 are shown in shaded boxes.

Domain	Physical		Psychosocial	
	This study	USA	This study	USA
physical functioning	0.889	0.82	0.077	0.08
role/social limitation emotional	0.782	0.20	0.305	0.72
role/social limitation physical	0.857	0.78	0.173	0.13
bodily pain	0.368	0.63	0.535	0.11
behaviour	0.600	-0.02	0.274	0.82
mental health	0.648	0.07	0.397	0.78
self esteem	0.140	0.03	0.752	0.75
general health perceptions	0.589	0.67	0.279	0.12
Parental impact time	0.515	0.41	0.616	0.69
parental impact emotion	0.150	0.36	0.835	0.75

Table 5.4 CHQ domain and summary scores

Table showing the mean and standard deviations for the CHQ domain and summary scores, for comparison with published norms [56].

Domain	Mean	Standard deviation
Physical Functioning	89.89	20.31
Role/Social Limitations Emotional	86.24	27.63
Role/Social Limitations Physical	86.42	27.07
Bodily Pain	76.48	26.35
Behaviour	65.42	20.35
Mental Health	73.74	17.40
Self Esteem	81.66	20.41
General Health Perceptions	62.25	19.41
Parental Impact Emotion	76.86	21.26
Parental Impact Time	85.52	23.68
Family Activities	74.25	23.86
Family Cohesion	77.72	20.28
Physical summary score	48.89	11.08
Psychosocial summary score	49.22	10.34

Table 5.5 Correlations between CHQ domains and frequency of pyrexia

Table showing the correlation between CHQ domain scores and the average number of days spent with pyrexial illness per month in children with recurrent AOM or sore throats (n=64).

CHQ Domain	Spearman's rho	p
Global health	-0.479	<0.001*
Physical functioning	-0.419	0.001*
Role/social limitations emotional	-0.259	0.039*
Role/social limitations physical	-0.364	0.003*
Bodily pain	-0.372	0.003*
Behaviour	-0.256	0.044*
Global behaviour	-0.028	0.831
Mental health	-0.330	0.008*
Self esteem	-0.371	0.003*
General health perceptions	-0.245	0.060
Change in health	-0.455	<0.001*
Parent impact emotion	-0.345	0.007*
Parent impact time	-0.249	0.053
Family activities	-0.400	0.001*
Family cohesion	-0.086	0.515

Table 5.6 TACQOL domain scores

The TACQOL domain scores, with mean and standard deviations for comparison with published norms [24].

Domain	Minimum	Maximum	Mean	Standard deviation
Body	8	32	22.4	6.17
Motor	11	32	30.1	4.09
Autonomy	3	32	30.1	4.55
Cognition	13	32	27.3	4.96
Social	15	32	29.7	3.18
Positive emotions	7	16	14.0	2.38
Negative emotions	3	16	10.5	3.07

Table 5.7 Internal consistencies of the TACQOL domain scores

Cronbach's alpha for the items in each of the TACQOL domains, not including the qualifying statements regarding emotional response.

Domain	Number of items	Alpha
Body	9	0.848
Motor	8	0.875
Autonomy	8	0.921
Cognition	8	0.862
Social	8	0.637
Positive emotions	8	0.826
Negative emotions	8	0.787

Table 5.8 TACQOL domain scores: age, sex and deprivation

The effect of age, sex and socio-economic deprivation on TACQOL domain scores (*Jonckheere-Terpstra test across 9 groups representing age in years or 5 groups representing the quintiles of the Carstairs Deprivation Index distribution; **Mann-Whitney test).

Domain	Age of child *	Sex of child **	Sex of respondent **	Carstairs Deprivation Index *	Manual vs Non-manual occupation **
Body	0.866	0.550	0.301	0.764	0.558
Motor	**0.039**	0.816	0.486	**0.010**	0.939
Autonomy	0.874	0.808	0.730	0.151	0.518
Cognition	0.891	0.514	0.145	0.400	0.188
Social	0.144	0.295	0.786	0.619	0.195
Positive emotions	0.359	0.901	0.358	0.537	0.976
Negative emotions	0.322	0.420	0.280	0.892	0.722

Table 5.9 TACQOL domain scores and overall HRQOL

All of the TACQOL domain scores were associated with overall HRQOL rated directly on a 100mm visual analogue scale or on a 5-point Likert rating scale, with 5 representing "poor" and 1 "excellent" quality of life (*Jonckheere-Terpstra test across 5 groups; **Spearman's rho).

Domain	p for Likert scale*	rho for visual analogue scale**	p for visual analogue scale
Body	<0.001	-0.567	<0.001
Motor	<0.001	-0.492	<0.001
Autonomy	<0.001	-0.406	<0.001
Cognition	0.001	-0.401	<0.001
Social	0.001	-0.277	0.018
Positive emotions	0.005	-0.393	0.001
Negative emotions	0.004	-0.312	0.008

Table 5.10 Internal consistencies of the TAPQOL domain scores

Cronbach's alpha for the items in each of the TAPQOL domains, not including the qualifying statements regarding emotional response.

Domain	Number of items	Alpha
Abdominal	3	0.419
Skin	3	0.762
Respiratory	3	0.614
Sleep	4	0.868
Appetite	3	0.880
Eating	3	0.583
Vitality	3	0.861
Positive mood	3	0.949
Aggression	7	0.883
Anxiety	3	0.708
Social behaviour	3	0.785
Motor	4	0.896
Communication	4	0.862

Table 5.11 TAPQOL domain scores: age, sex and deprivation

The effect of age, sex and socio-economic deprivation on TAPQOL domain scores (*Jonckheere-Terpstra test across 5 groups representing age in years or quintiles of Carstairs Deprivation Index distribution; **Mann-Whitney test; n=150)

Domain	Age of child *	Sex of child **	Sex of respondent **	Carstairs Deprivation Index *	Manual vs Non-manual occupation **
Abdominal	0.158	0.273	0.171	0.680	0.728
Skin	0.623	0.602	0.894	0.626	0.745
Respiratory	0.063	0.605	0.567	0.451	0.915
Sleep	0.149	0.308	0.555	0.830	0.973
Appetite	0.576	0.855	0.231	0.748	0.669
Eating	0.902	0.697	0.160	0.873	0.562
Vitality	0.023	0.067	0.926	0.449	0.316
Positive mood	0.797	0.984	0.161	0.083	0.056
Aggression	0.651	0.247	0.025	0.866	0.326
Anxiety	0.243	0.388	0.435	0.707	0.312
Social behaviour	0.105	0.691	0.928	0.212	0.420
Motor	0.121	0.000	0.492	0.244	0.387
Communication	0.679	0.281	0.913	0.694	0.588

Table 5.12 TAPQOL domain scores and referral diagnosis

Mean TAPQOL domain scores (standard deviations in brackets) for each diagnostic group, and for the whole study group.

Domain	OME n=67	rAOM n=47	Sore throats n=36	Whole group n=150
Abdominal	81.5 (±18.4)	84.8 (±16.1)	76.3 (±16.6)	81.4 (± 17.4)
Skin	87.3 (±17.7)	90.8 (±14.8)	83.1 (±21.9)	87.4 (±18.0)
Respiratory	94.0 (±12.8)	92.4 (±13.9)	87.4 (±19.7)	92.0 (±15.1)
Sleep	73.2 (±21.0)	61.2 (±22.0)	64.3 (±24.7)	67.2 (±22.7)
Appetite	84.2 (±15.3)	73.2 (±22.2)	53.2 (±29.6)	73.3 (±24.8)
Eating	92.7 (±11.3)	90.6 (±12.7)	69.6 (±27.7)	86.7 (±19.2)
Vitality	96.8 (±11.3)	91.5 (±19.9)	81.0 (±24.3)	91.3 (±18.9)
Positive mood	90.8 (±18.0)	89.4 (±19.5)	84.7 (±23.4)	88.9 (±19.8)
Aggression	61.4 (±25.8)	62.6 (±22.3)	63.5 (±21.0)	62.3 (±23.5)
Anxiety	75.5 (±22.1)	77.3 (±20.7)	79.6 (±22.2)	77.0 (±21.6)
Social behaviour	91.0 (±15.7)	89.7 (±19.5)	90.7 (±18.4)	90.5 (±17.4)
Motor	98.5 (±4.7)	98.5 (±4.1)	95.7 (±14.5)	97.8 (±8.3)
Communication	83.7 (±16.4)	87.3 (±14.5)	89.5 (±18.4)	86.3 (±16.5)

Table 5.13 TAPQOL domain scores and frequency of pyrexia

Table to show the TAPQOL domain scores that were correlated with average number of days pyrexia per month in the children with recurrent AOM or sore throats (n=105).

TAPQOL domain	Spearman's rho	p
Appetite	-0.465	<0.001
Eating problems	-0.435	<0.001
Sleep	-0.384	<0.001
Vitality	-0.318	0.001
Positive mood	-0.271	0.005
Abdominal problems	-0.246	0.013
Aggression	-0.236	0.016

Table 5.14 TAPQOL domain scores and frequency of sore throat

Table to show the TAPQOL domain scores that were correlated with the number of sore throats in the last year in the children with recurrent sore throats (n=37).

TAPQOL domain	Spearman's rho	p
Social behaviour	-0.545	<0.001
Abdominal problems	-0.528	0.001
Aggression	-0.457	0.004
Eating problems	-0.453	0.006
Sleep	-0.428	0.009
Motor functioning	-0.357	0.03
Vitality	-0.346	0.036
Respiratory problems	-0.360	0.037

Table 5.15 TAPQOL domain scores and OM6

Table to show the TAPQOL domain scores that were correlated with ear-related handicap measured by OM6 in the children with recurrent AOM or OME (n=110).

TAPQOL domain	Spearman's rho	P
Communication	-0.384	<0.001
Positive mood	-0.372	<0.001
Sleep	-0.339	<0.001
Aggression	-0.347	<0.001
Appetite	-0.300	0.002
Social behaviour	-0.256	0.011
Motor functioning	-0.237	0.018
Eating problems	-0.223	0.020
Anxiety	-0.216	0.024

Table 5.16 TAPQOL domain scores and HUI mark III

Table to show the TAPQOL domain scores that were correlated with overall health-related quality of life measured using the Health Utilities Index mark III (n=122).

TAPQOL domain	Spearman's rho	P
Communication	0.582	<0.001
social behaviour	0.381	<0.001
Positive mood	0.360	<0.001
Appetite	0.332	<0.001
Motor functioning	0.324	<0.001
Sleep	0.312	0.001
Aggression	0.285	0.002
Anxiety	0.188	0.038

6 The MRC Quality of Family Life Questionnaire

6.1 Background

Living with a person who has an illness has an impact on the rest of the family. Decisions in paediatric health care are determined by the family as much as by the individual. The decision to seek health care intervention, and the nature of the intervention chosen, may reflect the impact the disease has on the family more than the impact of the disease on the individual. The extent to which such decisions are influenced by the child's perceived quality of life as opposed to the impact on the family is currently unknown, but there is evidence that parental factors play a large part in the decision to seek medical attention [61]. Teasing out such influences will require instruments to measure child and family impact separately.

Currently, there are few instruments available to assess family impact. Instruments do exist to measure the family impact of specific conditions such as asthma [113], developmental disabilities [114] and ear, nose and throat infections [109]. The Child Health Questionnaire [56], in contrast, is generic in scope and includes questions in four of its domains which come under the heading of quality of family life. While this makes it one of the few generic instruments to assess family impact, the benefit of this is lost when the child- and family-centred information is combined in the two summary scores produced (Physical and Psychosocial).

The Medical Research Council Institute for Hearing Research has produced a Quality of Family Life questionnaire (QOFL) which is intended to be generic in scope (see appendix). An initial list of potential questions was produced by "brainstorming" within the group, and the 55 questions produced were then reduced to 26 after piloting on the family members of the group and 4 parents of children who had undergone ventilation tube insertion for otitis media with effusion. The questionnaire has been used in a clinical study on the effect on the family of tinnitus in adults, although four extra communication-specific questions were added for this project [115]. These initial results suggest a six-factor structure (day to day activity with patient, effect on patient, effect on family, coping, understanding, restriction of activities) and that the questionnaire can discriminate those who have been seen at a specialist tinnitus clinic from those awaiting an appointment. Further validation of the original version was planned, using data from 23 adult cochlear implant recipients, 11 adults who have undergone middle ear surgery, 20 parents of children who have received cochlear implants and 15 parents of children with hearing aids (H Fortnum, personal communication). However, this process was never completed and the development of the instrument was taken no further.

The QOFL is particularly interesting in that its theoretical basis is novel: the instrument is designed to assess the impact of a condition on "the family" as a functional unit, rather than on any individual within it. To ask a parent "what impact does your child's disease have on your quality of life?" is quite distinct

from "what impact does your child's disease have on your family's ability to function?"

6.2 Study aims

The purpose of this study was to use the QOFL to assess the impact of common childhood otolaryngological conditions (sore throats, otitis media) on the families of the affected children. In doing so, we would be able to test the appropriateness of the QOFL for use in this context by studying the extent to which the impact on the family is related to the severity of the child's disease and the child's own perceived quality of life.

These conditions were chosen as the most common medical conditions of childhood and the most common reason for parents to seek medical attention for their child. The high level of demand from medical services suggests significant impact on the families from these conditions.

Our hypothesis was that a valid measure of quality of family life would show that, on average, more severe disease in a child would produce a greater impact on the family. Thus we would expect an association between markers of disease severity and QOFL scores. We would also expect a greater association between QOFL and the family- and parent-orientated domains of the CHQ than

the other (child-orientated) domains. Ideally, the QOFL should be free from the influence of extraneous variables such as age, sex and socio-economic class.

6.3 Participants and methods

Prior approval for the study was obtained from local research ethics committees.

A consecutive series of children was recruited for the study from the paediatric otolaryngology clinics of three hospitals in the West of Scotland (Crosshouse Hospital, Kilmarnock; Ayr Hospital; The Royal Hospital for Sick Children, Yorkhill, Glasgow). To be eligible for inclusion, the children had to be at their first hospital visit after being referred by their General Practitioner with suspected otitis media with effusion (OME), recurrent acute otitis media (AOM) or sore throats.

At the time of their hospital visit, the parents were asked to complete the QOFL. Parents also completed various quality of life measures on behalf of the child at the same time. These included the Child Health Questionnaire (CHQ, 50-question parent form) for those children aged 5 years or older [56] and global ratings of the child's quality of life on a 10cm visual analogue scale and a 5-point Likert-type rating scale. The otitis media-related handicap measure OM6 [93] was also completed when relevant. All children then underwent a standard

clinical consultation with the same otolaryngologist (HK) where clinical data were collected. Written parental consent for study participation was obtained in every case.

Data were stored on a computer, and statistical analyses were performed using SPSS version 11.0. Factor analysis was performed using a principle component analysis, selecting for eigenvalues greater than 1, and using varimax rotation.

6.4 Results

274 children were seen in the clinics. Twenty-three declined to participate in this study, leaving 251 children with data for analysis. 129 were boys, 122 girls, and the median age was 5 (range 1-14 years). The primary referral diagnosis was OME in 123 cases, recurrent AOM in 57 and sore throats in 71. In many cases, however, more than one symptom was present on enquiry. The same cohort of children was used for this study as for the studies described in Chapter 5, although failure to complete some questionnaires meant that overlap between the studies was not complete (see study recruitment flow diagram, Appendix 2).

Everybody's definition of what constitutes a family will be different, so the questionnaire allows the respondent to make the judgement about who to include. Guidance is given as, "your immediate family, usually people who

live with you." Space is given for the family members to be listed, along with their relationship to the child. For the purposes of the study, no distinction was drawn between parents, adoptive parents, step-parents and unmarried partners. In most cases, the family was based on a traditional mother-father couple. In 48 families (20%), there was only a single parent included (45 mothers, 3 fathers). The fathers were aged between 19 and 59 years (median 36) and the mothers 19-53 (median 34). Fourteen families included other adults, mostly grandparents. 173 (72%) of the children had siblings, between 1 and 5 in number, and 2 families included other children, in both cases the siblings of the parents.

None of the parents had any difficulties with the concepts or wording of the questions in the QOFL. The 251 completed QOFL questionnaires contained a total of 6526 items, of which 137 (2.1%) were left uncompleted.

The QOFL scores were not associated with the age of the child (Figure 6.1), the sex of the child (Figure 6.2), the sex of the respondent (mother versus father, Figure 6.3), or the degree of socio-economic deprivation (assessed from the postcode area of residence using the Carstairs Deprivation Index [111], Figure 6.4), but scores were slightly lower in those families where the main wage earner was in a manual rather than non-manual occupation (Mann-Whitney, p=0.016, Figure 6.5). When separated by the primary reason for referral, worse QOFL scores were seen in the sore throats group than the OME group, with the recurrent AOM group intermediate between the two (Figure 6.6).

To look for an effect of disease severity on QOFL scores, the children were divided into groups according to the symptoms present on enquiry in the clinic (which occasionally differed from the referral diagnosis). In the 160 children with recurrent pyrexial illnesses (AOM or sore throats), the QOFL score was inversely correlated with the average number of days pyrexia per month (Spearman's rho=-0.297, p<0.001, Figure 6.7). The QOFL scores were also worse in those who had needed to take time off school in the last year because of their ear or throat infection, compared with those who had not (Mann-Whitney, p=0.014, Figure 6.8).

Within the sore throats group (74 children), QOFL scores were only weakly inversely correlated with the number of sore throats in the last year (Spearman's rho=-0.225, p=0.054, Figure 6.9). Within the recurrent AOM group, however, QOFL scores were not associated with either frequency of otalgia or otorrhoea (Spearman's rho=-0.129 and -0.093 respectively, Figure 6.10 and Figure 6.11). Within the OME group, there was no association between QOFL scores and the presence of hearing thresholds of 25dBHL or worse in the better ear (Mann-Whitney, p=0.652, Figure 6.12), or with bilateral B/C2 tympanograms (Mann-Whitney, p=0.67, Figure 6.13).

For the group of children with recurrent AOM or OME, ear-specific handicap rated on OM6 was correlated with QOFL score (Spearman's rho=-0.423, p<0.001, Figure 6.14). The child's overall quality of life was rated directly by

parents on a 10cm visual analogue scale and a 5-point Likert rating scale. Both correlated with the QOFL score (Spearman's rho=-0.346, p<0.001 for the visual analogue scale, Figure 6.15; Jonckheere-Terpstra, p<0.001 for the 5-point scale, Figure 6.16).

The child's overall quality of life was also assessed with the CHQ. QOFL scores correlated with the CHQ Psychosocial summary score (Spearman's rho=0.594, p<0.001) and with the CHQ Physical summary score (rho=0.223, p=0.016, Figure 6.17). All 11 of the domain scales in the CHQ were significantly correlated with the QOFL, but the Family Activities scale was the most highly correlated (Table 6.1).

The items of the QOFL were found to be highly internally consistent, with a Cronbach's alpha of 0.91. Item-total correlations showed that alpha would only be increased when one item (question 23: "how much does your family understand about your child's ear, nose and throat problems?") was deleted (Table 6.2).

Factor analysis was performed using a principle component extraction with varimax rotation. Although this initially suggested that there were six factors in the data, two of these proved to be unstable when random 10% subsets of the data were deleted and the factor analysis repeated. This suggests that only four of the factors are true descriptors of the whole dataset, the other two being unduly influenced by the presence of certain key cases. Table 6.3 shows the

extent to which each question in the QOFL loads onto each of the four factors, which we have labelled for convenience as "Enjoyment Within the Family", "Coping and the Future", "Pressure and Restrictions", "Inclusion and Embarrassment".

Comparing the factor scores between children grouped by referral diagnosis, the only one which differed between groups was "Pressure and Restrictions" (mean scores 0.38 for OME, -0.15 for recurrent AOM, -0.5 for sore throats; one-way ANOVA*, $p<0.001$; Figure 6.18). The factor score "Pressure and restrictions" was the only one to be significantly associated with need to take time off school (t-test, $p<0.001$). None of the factor scores was associated with frequency of otalgia, frequency of otorrhoea, presence of B/C2 tympanograms or presence of a better ear threshold of 25dB or worse: only frequency of sore throat came close to having an association with "Pressure and Restrictions" (t-test, $p=0.088$; Figure 6.19).

6.5 Discussion

A difficulty in constructing a questionnaire of this kind is ensuring that it is appropriately worded for all the family types which it attempts to encompass. While a parent in a two-parent family with more than one child could be expected to conceptualise the construct of "family" and answer questions

* Non-parametric statistics are used throughout this thesis where the data are clearly skewed: the exception is the analysis of factor scores which have an approximately normal distribution.

appropriately, a single parent with only one young child may well have to give simply their own, personal perspective. Of course, in such a small family unit that would be an entirely appropriate response. None of the families surveyed in this study reported any difficulties with the concepts or wording in the QOFL.

Validity is something which cannot be proved outright in the absence of a "gold standard" measure of quality of family life for comparison. Correlation of the QOFL with comparable domains of the CHQ constitutes evidence of concurrent validity, meaning that both are trying to measure the same thing.

Other supporting evidence for validity, however, can be found by setting up and testing hypotheses about how a valid instrument would be expected to perform in various circumstances (construct validity). We have shown, for example, that where the disease has a greater impact on the child's HRQOL (however that is rated), it also impacts to a greater extent on the family. More severe disease, as estimated by frequency of sore throats and pyrexia, is also associated with a greater impact on the family, but the associations are weak. Audiometric thresholds and tympanometry show no association with family impact, but perhaps an association would be more likely with hearing difficulty as reported by the family, rather than objective measures, which are one step removed from the family's experience. Otorrhoea also does not seem to be associated with family impact, but then one could argue that an ear discharge of itself is unlikely to impact on other family members, other than by its smell, and

that the discharge encountered in AOM is usually very short-lived. The higher family impact of recurrent pyrexial illness as opposed to OME is not surprising when one considers time off work, sleepless nights and the emotional impact of caring for a child in pain [61].

The QOFL scores are not unduly influenced by extraneous factors such as age, sex and social class. Further evidence for its robustness comes from the completely different nature of the populations in which it has been used, and found to work successfully, namely adult carers of adults with tinnitus [115] and, now, parents of children with ear and throat infections.

Reliability, meaning here the precision of the measurement, is partly addressed by measures of internal consistency (such as Cronbach's alpha), and partly by measures of reproducibility. For the QOFL, the most interesting of these would be the extent to which two observers (mother and father, for example) agree in their responses about the same patient. This was not formally assessed in this study as only one questionnaire was completed for each child.

We have been able to show that "quality of family life" exists as an entity that parents can conceptualise and answer questions on, and that common ear and throat conditions have a measurable impact on it. The high internal consistency of the questionnaire would support the notion that the questions, although asking about a very varied range of issues, are tapping into a coherent concept of family life.

Although the high internal consistency shows that the questions all point broadly in the same direction, towards a coherent entity of "quality of family life", there is evidence from factor analysis that within this entity some of the questions group together more closely than others. Factor analysis is a procedure which aims to reduce a set of observations into a smaller number of sub-scores or factors to summarise the observations and give some indication of their underlying structure. The 26 questions from the QOFL were subjected to such an analysis. A direct comparison with the factor structure described in the previous study [115] was not possible due to the additional questions used in that study. Four factors were found within our data, which we have chosen to label "Enjoyment Within the Family", "Coping and the Future", "Pressure and Restrictions", "Inclusion and Embarrassment". The factor scores for "Pressure and Restrictions" differed according to referral diagnosis in a way that suggests that the disturbance that OME causes the family, unlike with sore throats and recurrent AOM, is largely determined by issues other than restriction of activities. This is understandable, given that child with recurrent pyrexial illness will often be too ill to participate in family activities, whereas the child with OME is not so restricted, but generates concern regarding speech, language, behaviour and education.

To summarise, quality of family life is a reasonable and useful entity to measure, and the QOFL measures it in a valid, robust and reliable way.

Figure 6.1 QOFL and child's age

Boxplot of child's age against QOFL scores (JonckheereTerpstra, p=0.292).

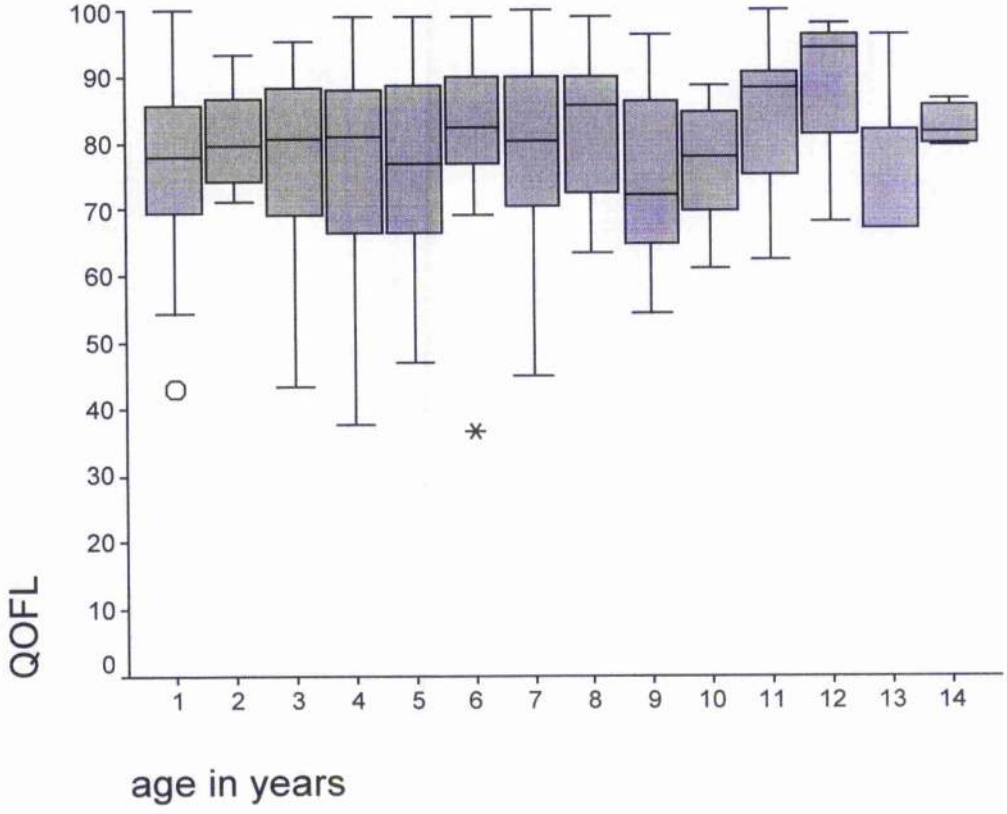


Figure 6.2 QOFL and sex of child

Boxplot of QOFL scores according to the sex of the child (MannWhitney, p=0.315).

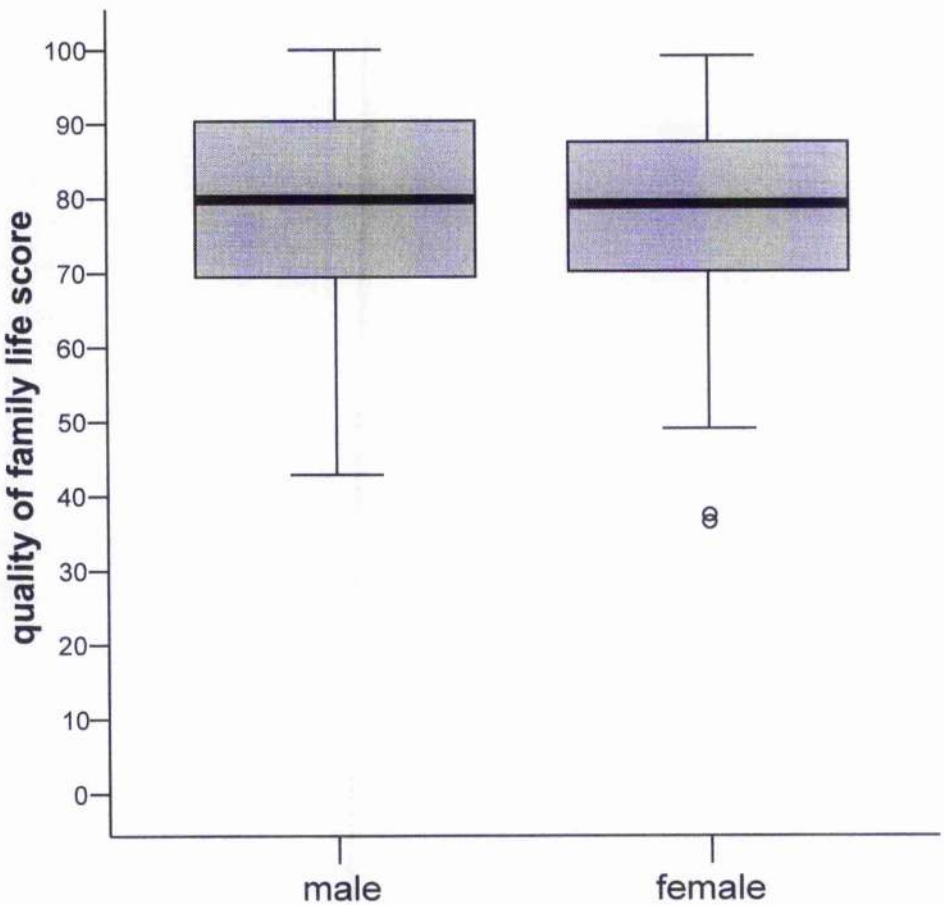


Figure 6.3 QOFL and sex of respondent

Boxplot of the sex of the person who filled in the forms against QOFL scores (Mann-Whitney, p=0.112). Three children whose forms were filled in by their grandmother were excluded from this analysis

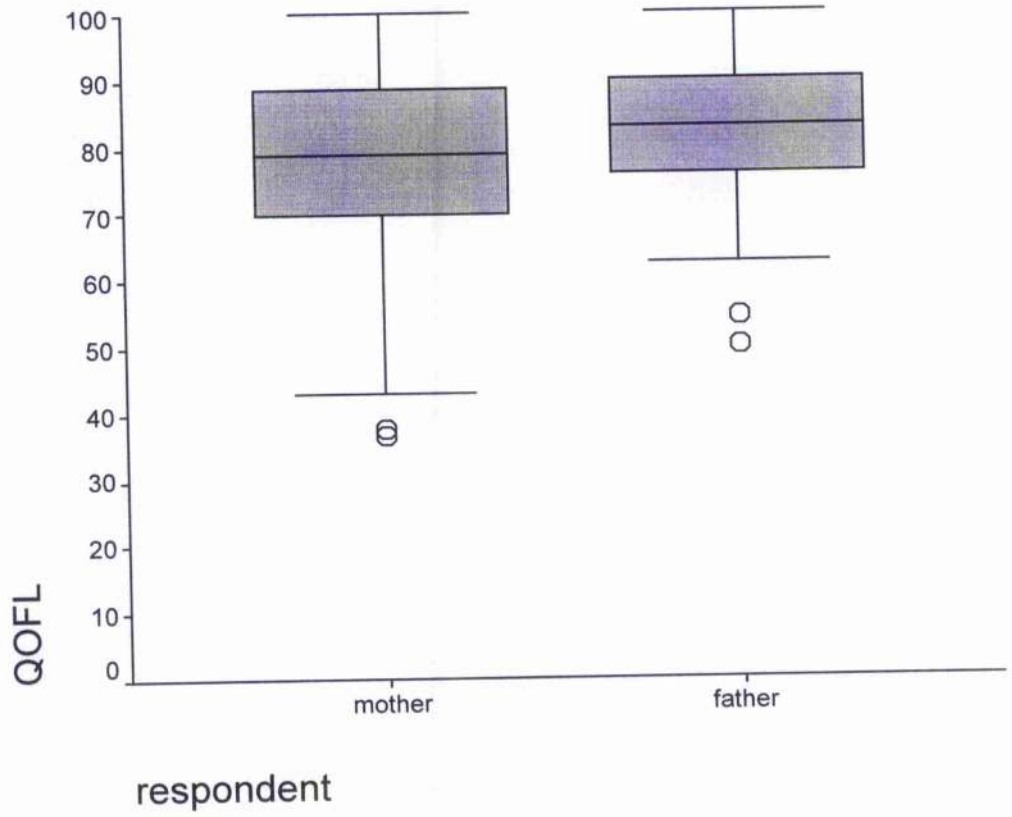


Figure 6.4 QOFL and socio-economic deprivation

Boxplot of the Cartairs Deprivation Index against QOFL score. The Carstairs Deprivation Index has been used to divide the children into 5 groups, with 1 being the most affluent and 5 the least affluent: the Index is structured such that 20% of the Scottish population falls into each of these groups (Jonckheere-Terpstra, p=0.459).

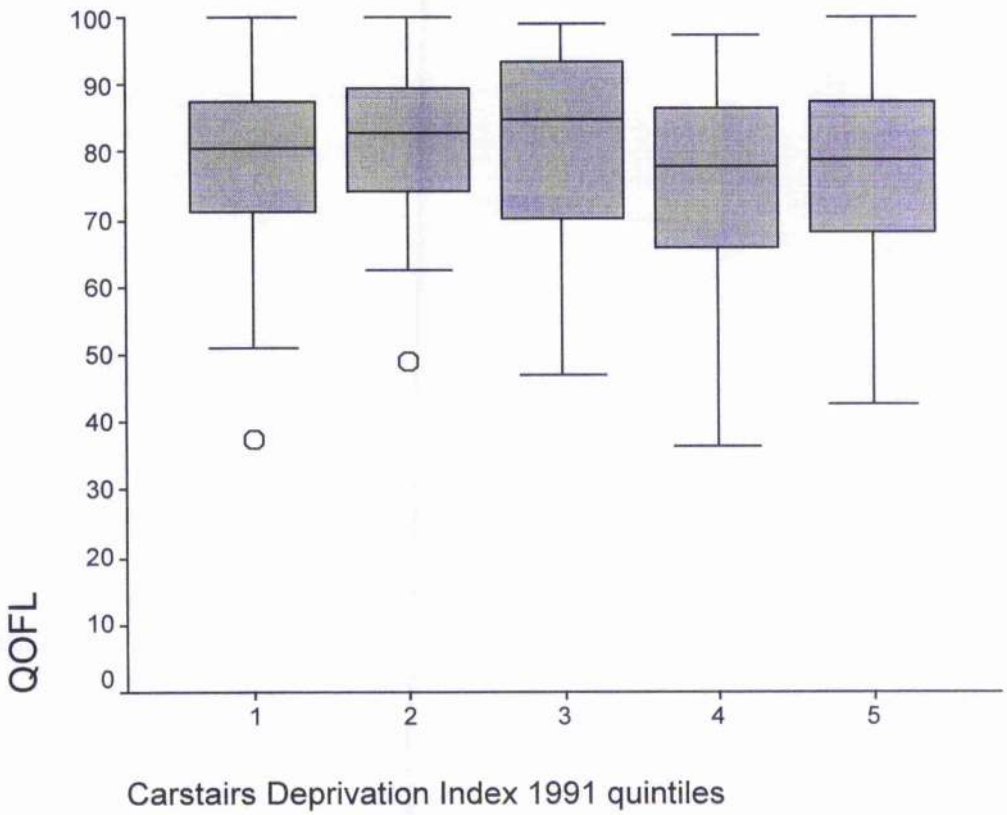


Figure 6.5 QOFL and parent's occupation

Boxplot of main wage-earner's occupation against QOFL score (Mann-Whitney, p=0.016).

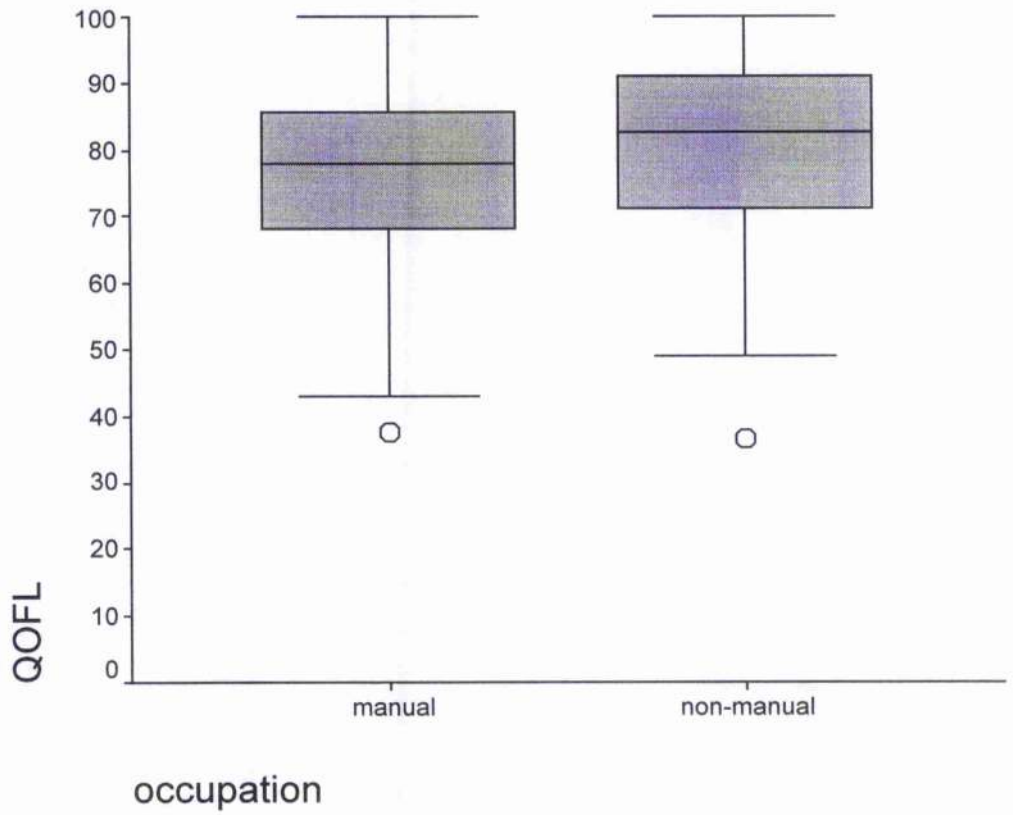


Figure 6.6 QOFL and referral diagnosis

Boxplot of primary referral diagnosis against QOFL score (Kruskal-Wallis, p=0.016)

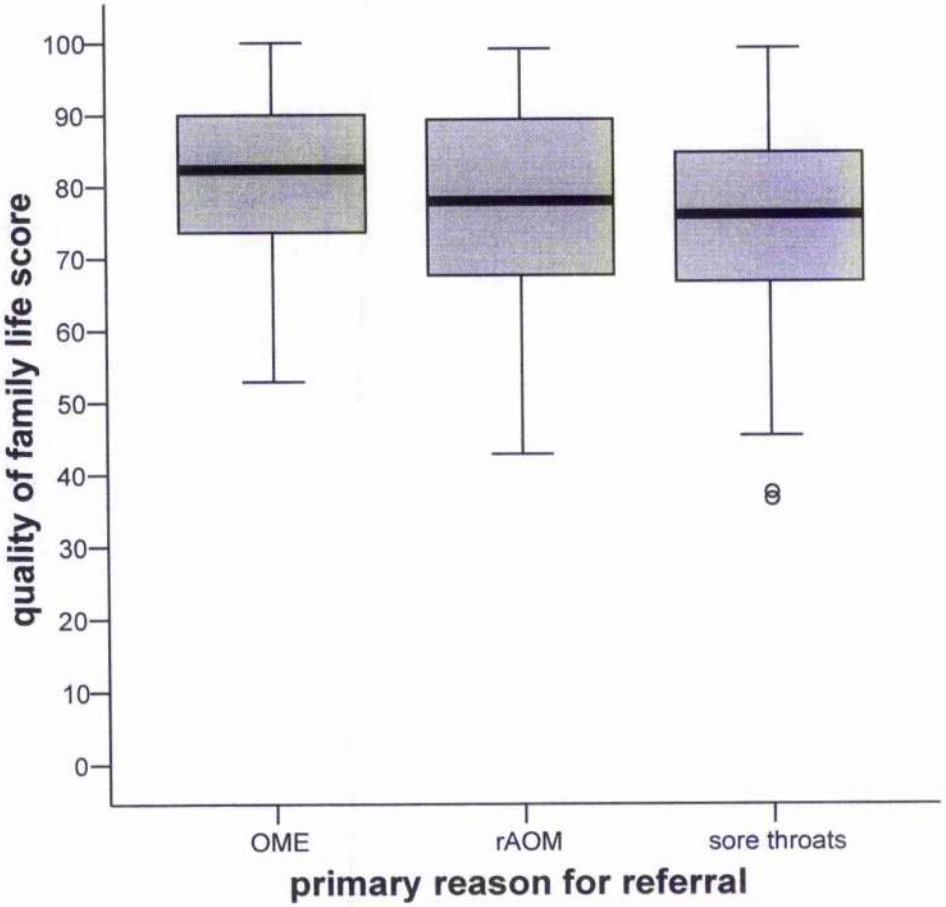


Figure 6.7 QOFL and pyrexial illness

Scatterplot showing the extent to which the QOFL score correlates with the average number of days per month that the child spends with pyrexia, for the children with sore throats or recurrent AOM only. The correlation is weak but significant (Spearman's rho= -0.297, $p<0.001$).

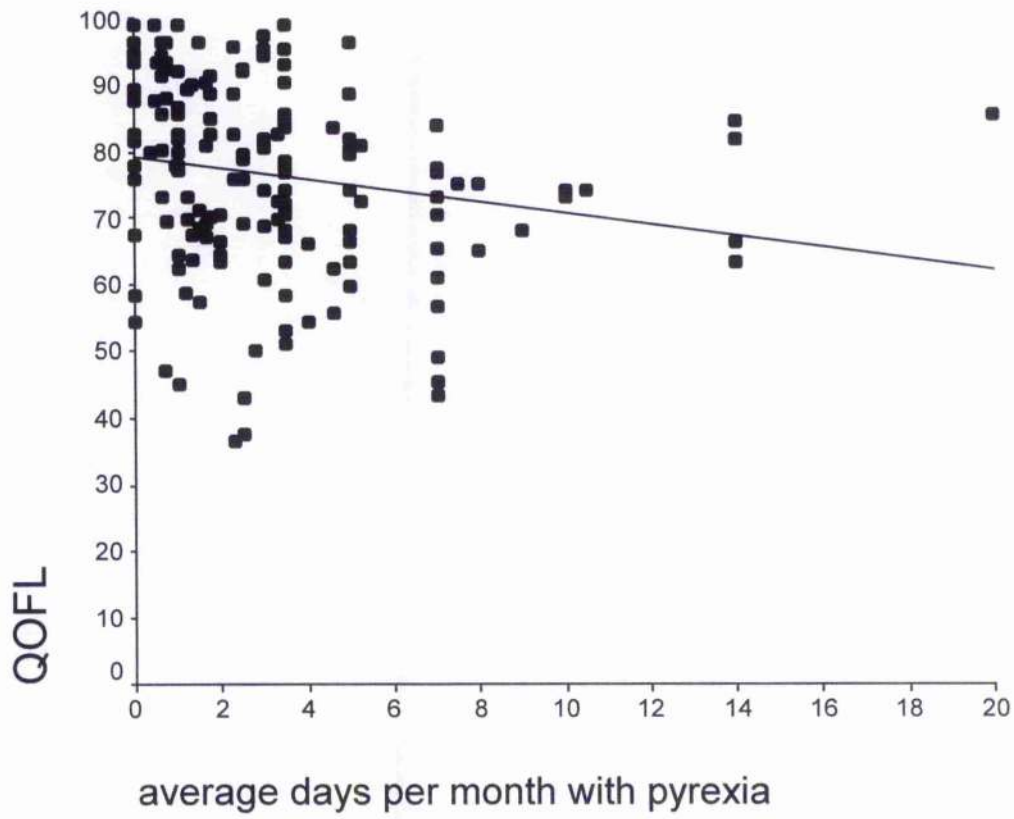


Figure 6.8 QOFL and time off school

Boxplot showing the QOFL scores for those children with recurrent AOM or sore throats who have needed time off school because of their illness, compared with those who have not (Mann-Whitney, p=0.014)

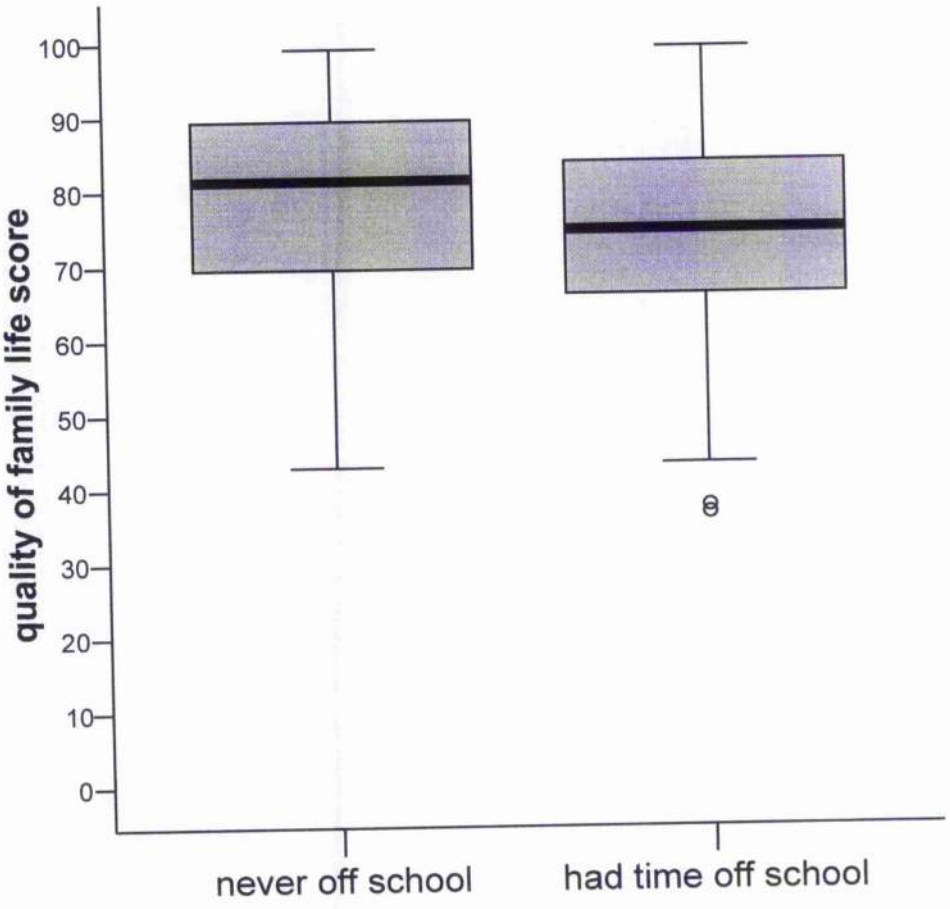


Figure 6.9 Correlation between QOFL and sore throats

Scatterplot of QOFL scores against number of sore throats in the last year. The correlation is weak (rho=-0.225, p=0.054).

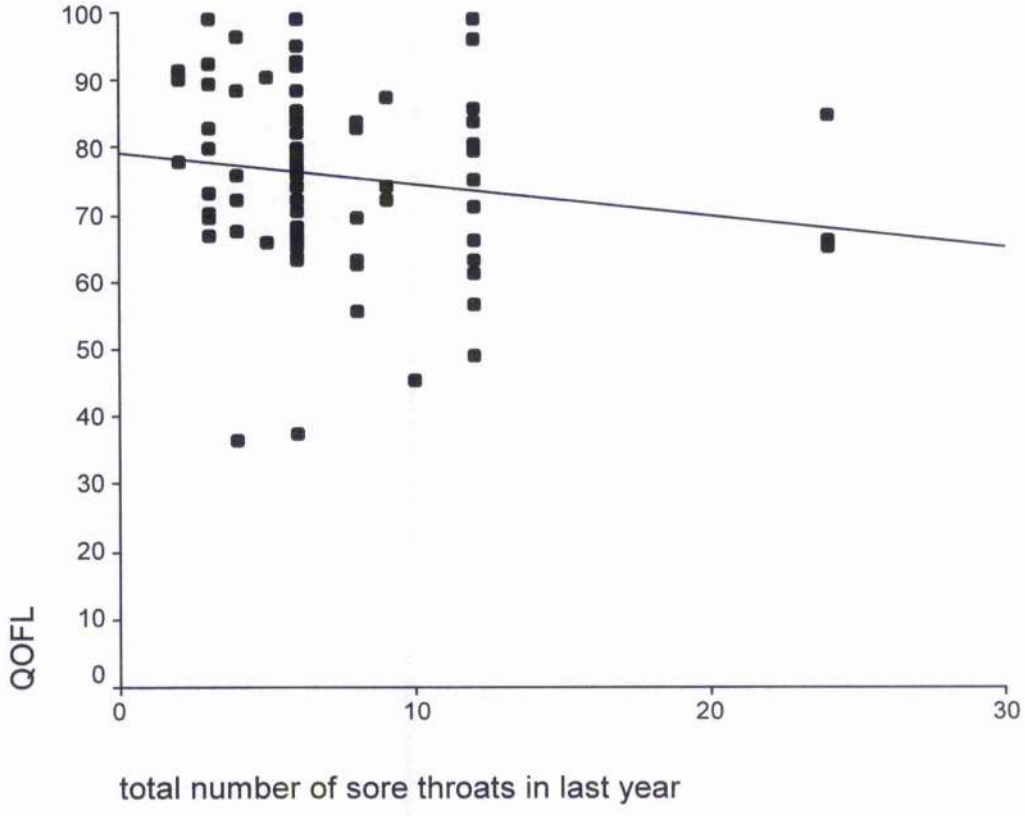


Figure 6.10 QOFL and frequency of ear infection

Boxplot showing slightly (but not significantly) worse QOFL scores in the families of children with at least one ear infection every 2 months compared with those with infections at a lesser frequency (Mann-Whitney, p=0.19). One infection every 2 months is the median frequency in this group.

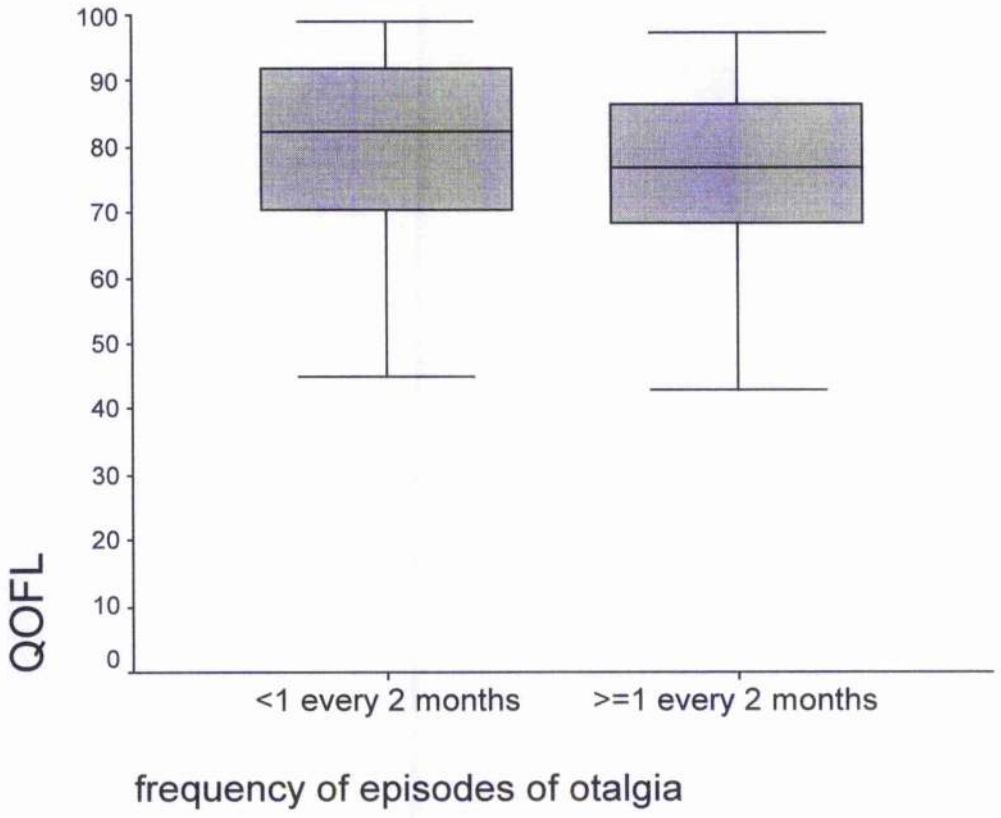
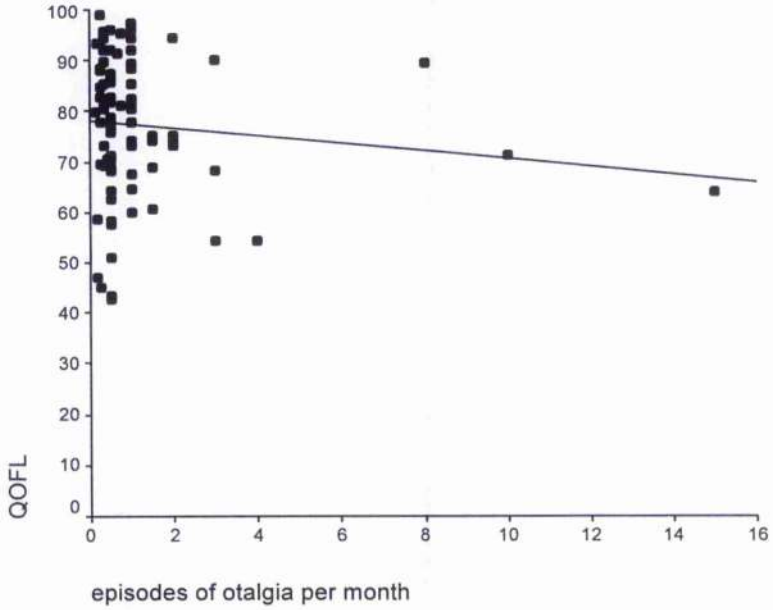


Figure 6.11 QOFL, otalgia and otorrhoea

Scatterplots showing a lack of correlation between frequency of otalgia and otorrhoea and QOFL scores in the children with ear infections (rho=-0.129 and -0.093 respectively)



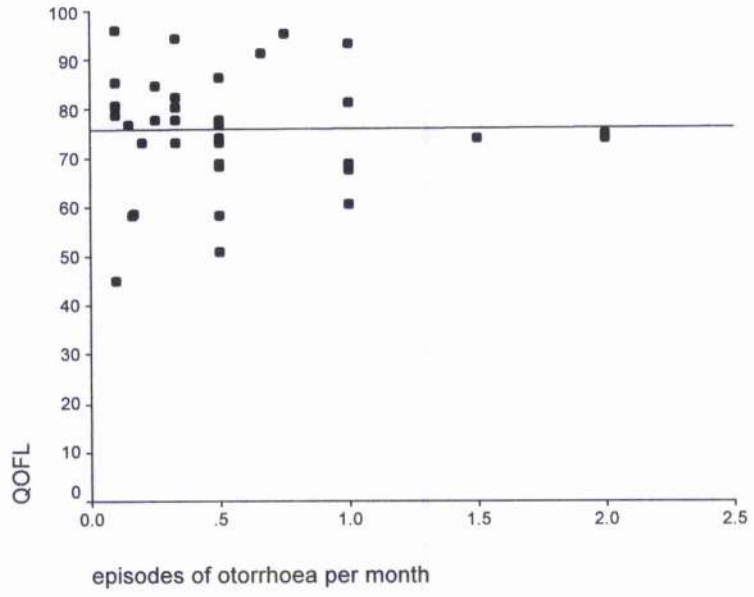


Figure 6.12 QOFL scores and hearing thresholds

Boxplot of QOFL scores in children with symptoms of OME according to their hearing thresholds at the clinic assessment. Children are grouped according to whether their better ear had a threshold of 25dBHL or worse (Mann-Whitney, p=0.652).

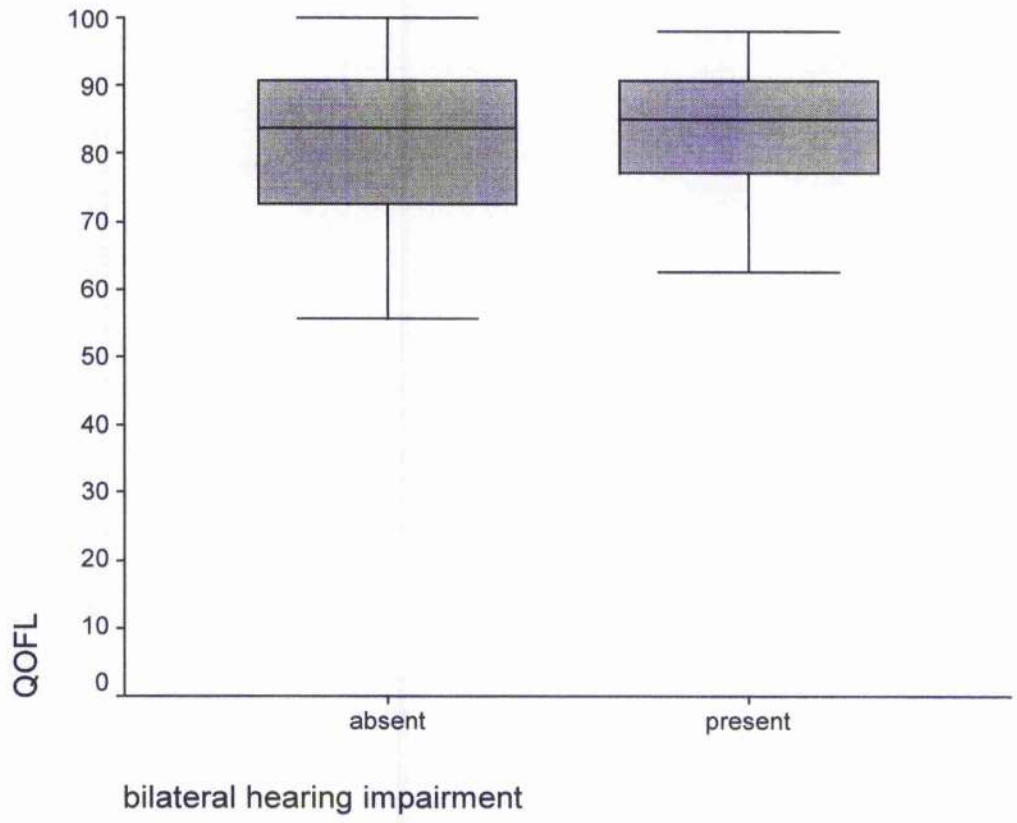


Figure 6.13 QOFL scores and tympanometry

Boxplot of QOFL scores in children with symptoms of OME according to the presence of bilateral middle ear effusions at the time of the clinic assessment, as shown by tympanometry (Mann-Whitney, p=0.669). Type B and C2 tympanograms are highly predictive of middle ear fluid, while type A and C1 tympanograms are highly predictive of a dry middle ear.

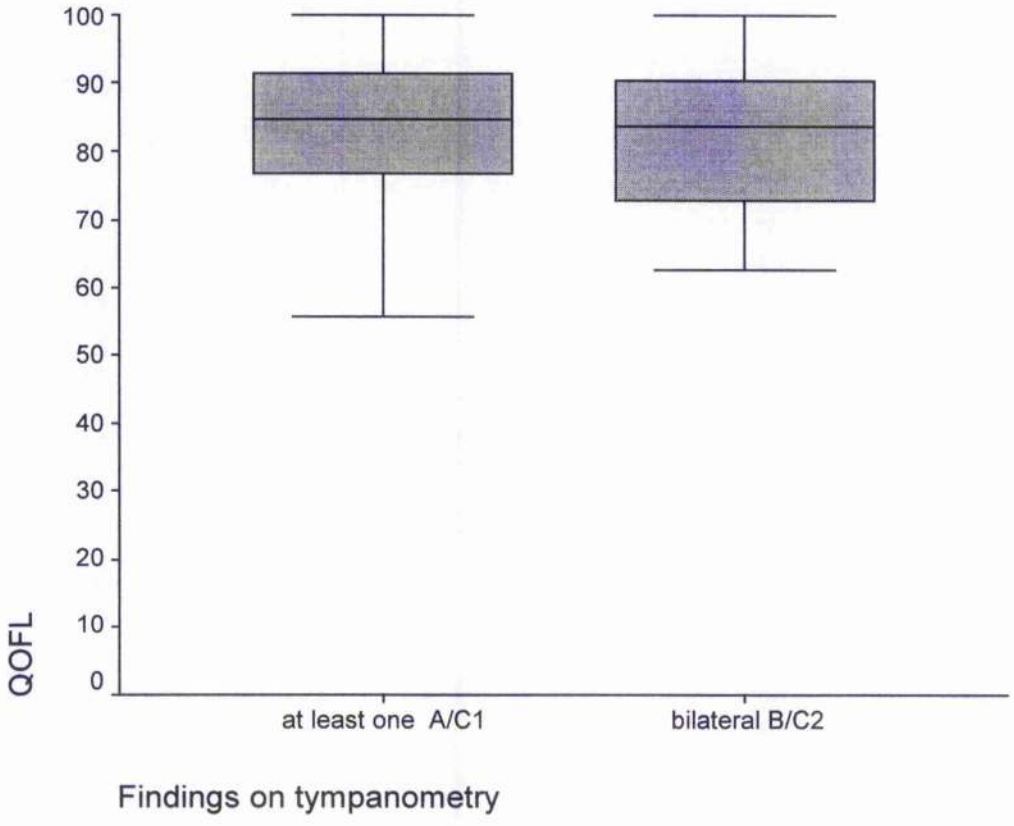


Figure 6.14 QOFL and OM6

Scatterplot of QOFL scores in the children with ear problems and ear-specific handicap as rated on OM6 (rho=-0.423, p<0.001).

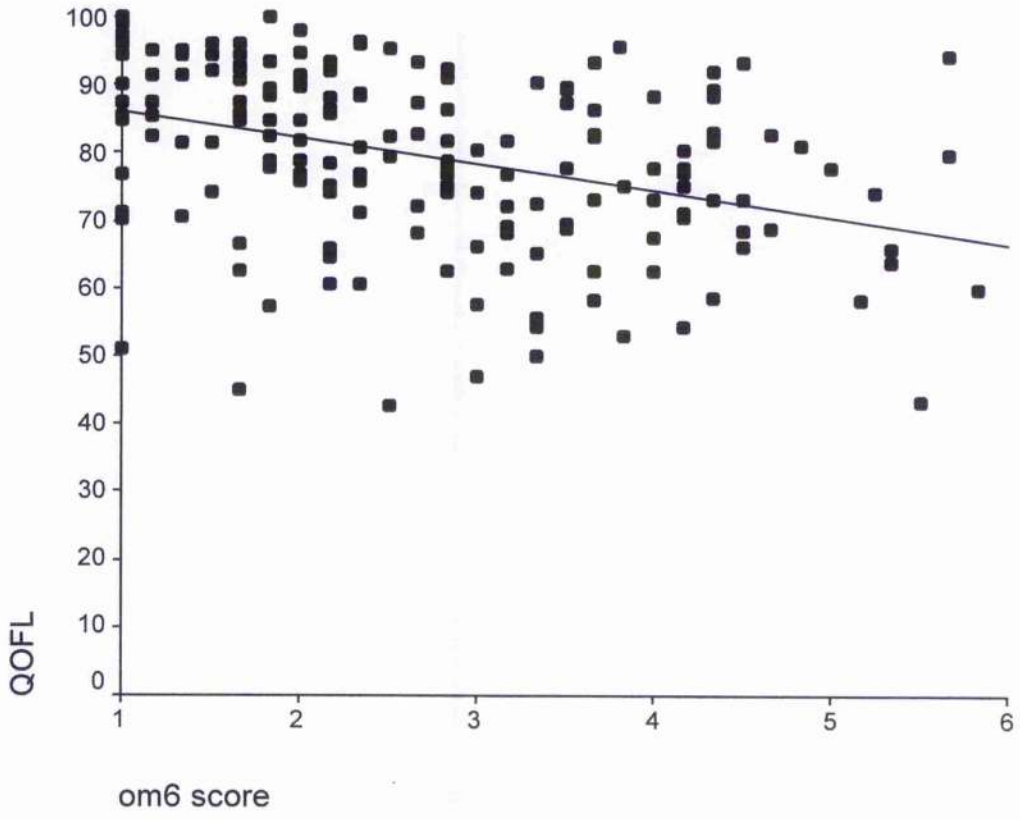


Figure 6.15 QOFL and HRQOL rated on a VAS

Scatterplot of QOFL scores and the parent's rating of the child's overall HRQOL as rated on a 100mm visual analogue scale (rho=-0.346, p<0.001).

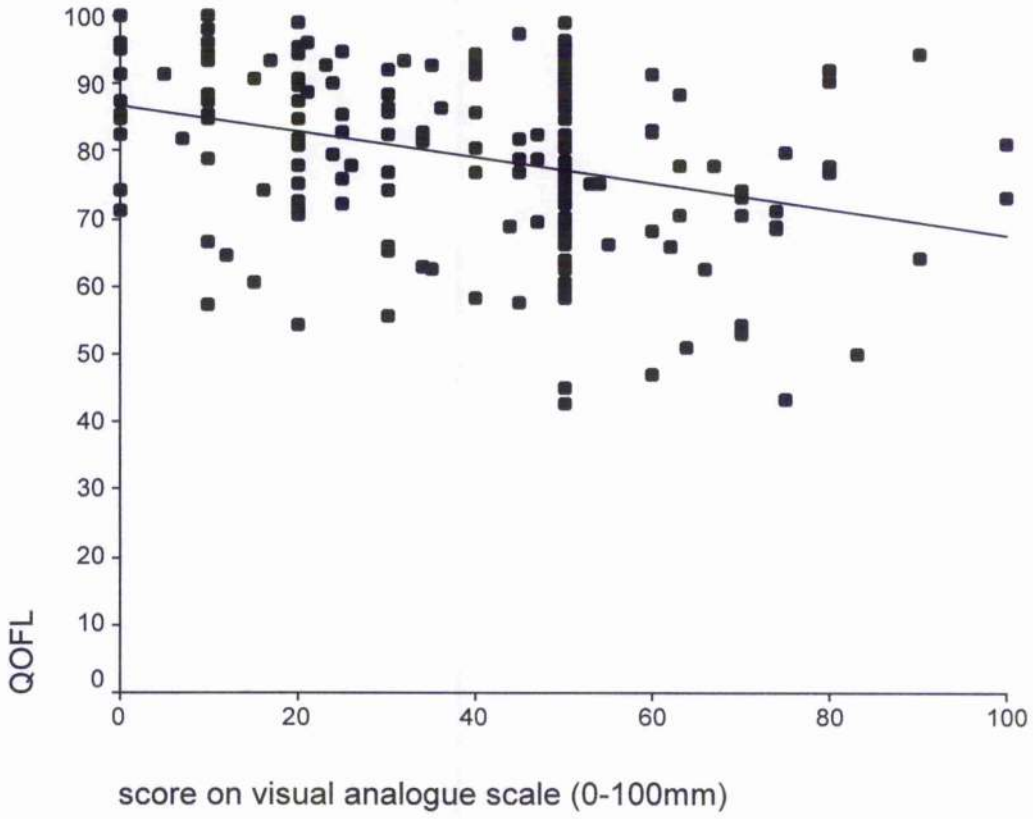


Figure 6.16 QOFL and HRQOL rated on a 5-point Likert scale

The child's overall health-related quality of life, as rated on a 5-point rating scale, and its association with the QOFL score (Jonckheere-Terpstra test, $p<0.001$). On the scale used, 1 represents "excellent" and 5 "poor" overall quality of life for the child.

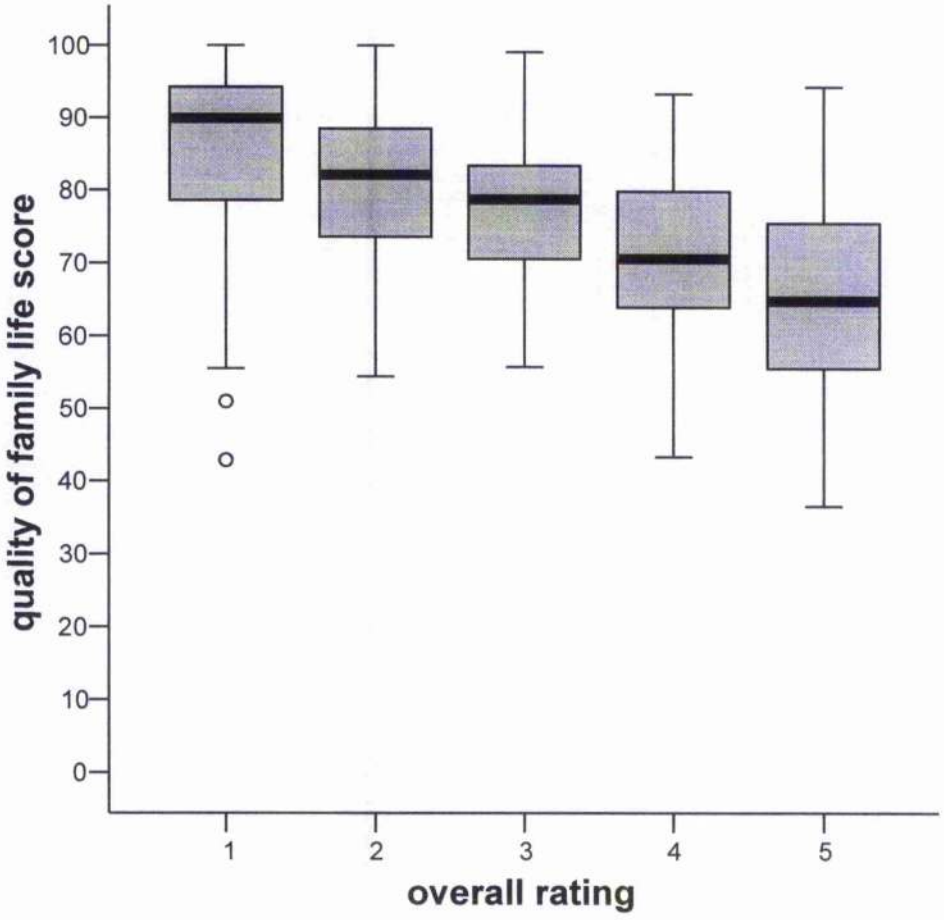
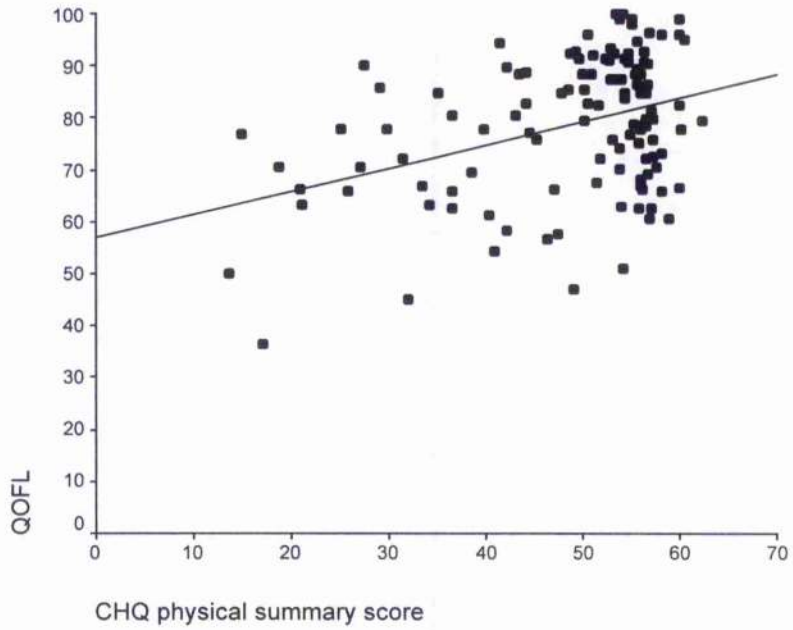


Figure 6.17 QOFL and CHQ summary scores

Scatterplots showing the correlation between the two summary scores of the CHQ (Physical and Psychosocial) and QOFL (rho for Physical=0.223, p=0.016; rho for Psychosocial=0.594, p<0.001).



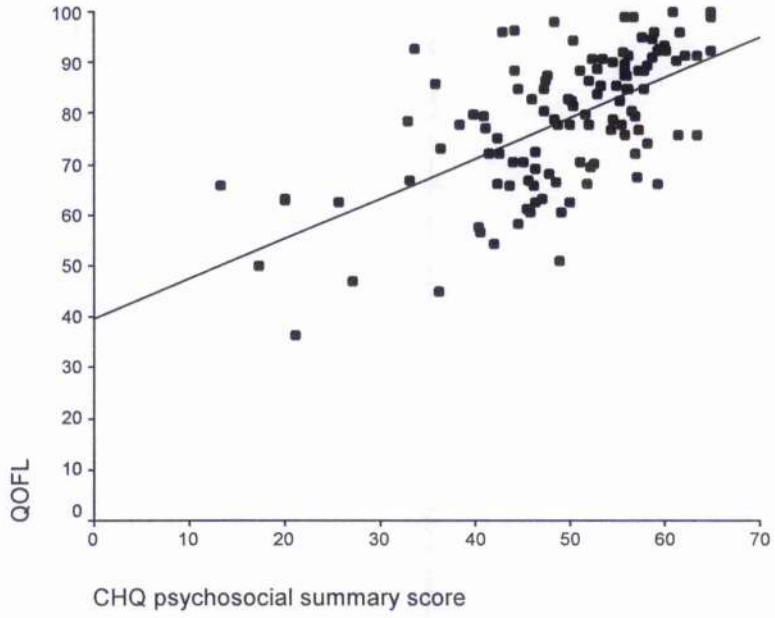


Figure 6.18 QOFL factor scores and primary reason for referral

Boxplots showing how the four factor scores relate to referral diagnosis. Only "Pressure and restrictions" is significantly different between the groups (one-way ANOVA, $p<0.001$).

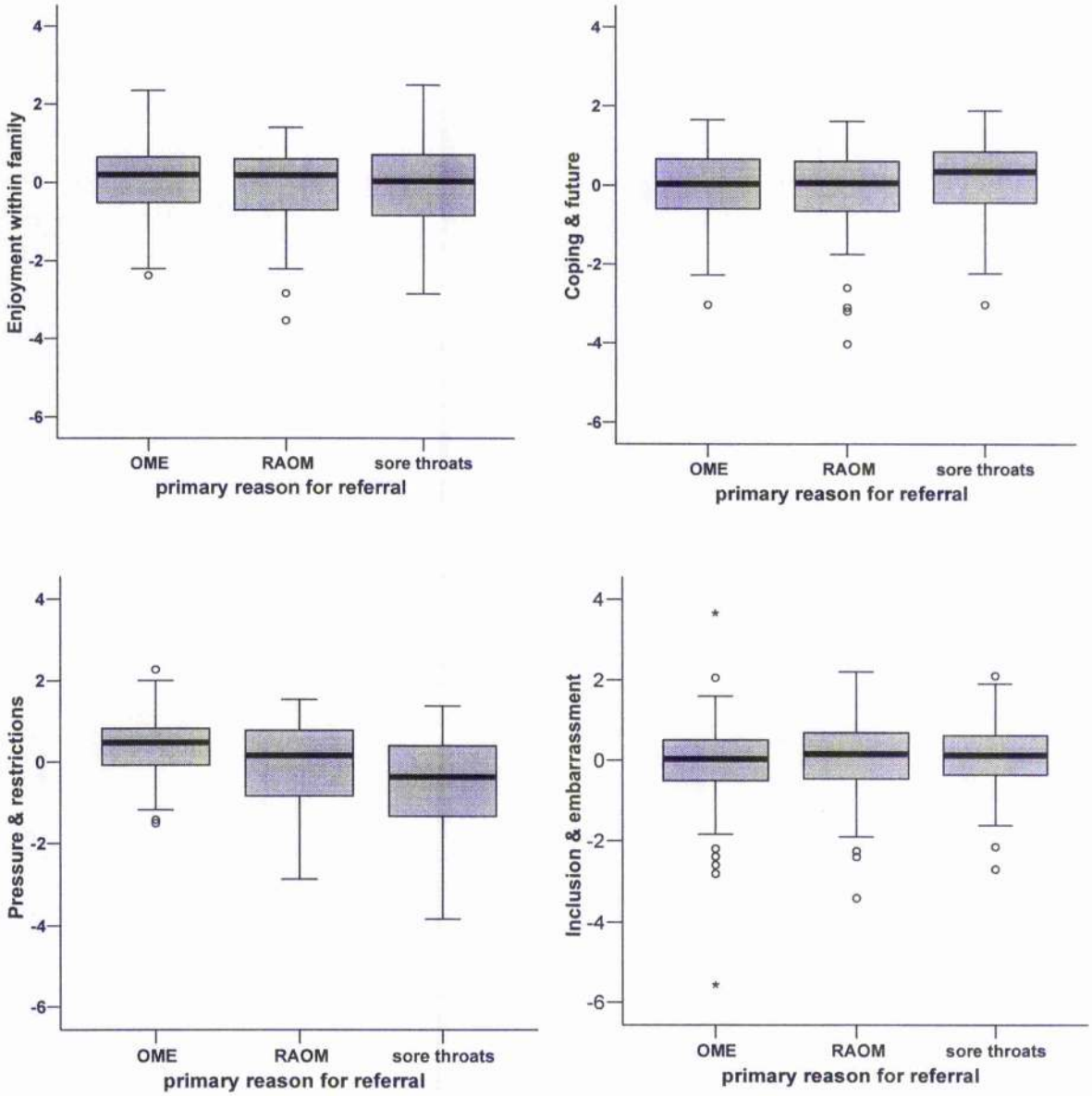


Figure 6.19 QOFL factor scores and frequency of sore throats

Boxplots to show the effect of freqency of sore throats on the four factor scores. Only "Pressure and restrictions" is significantly different between the two groups (t-test, p=0.088).

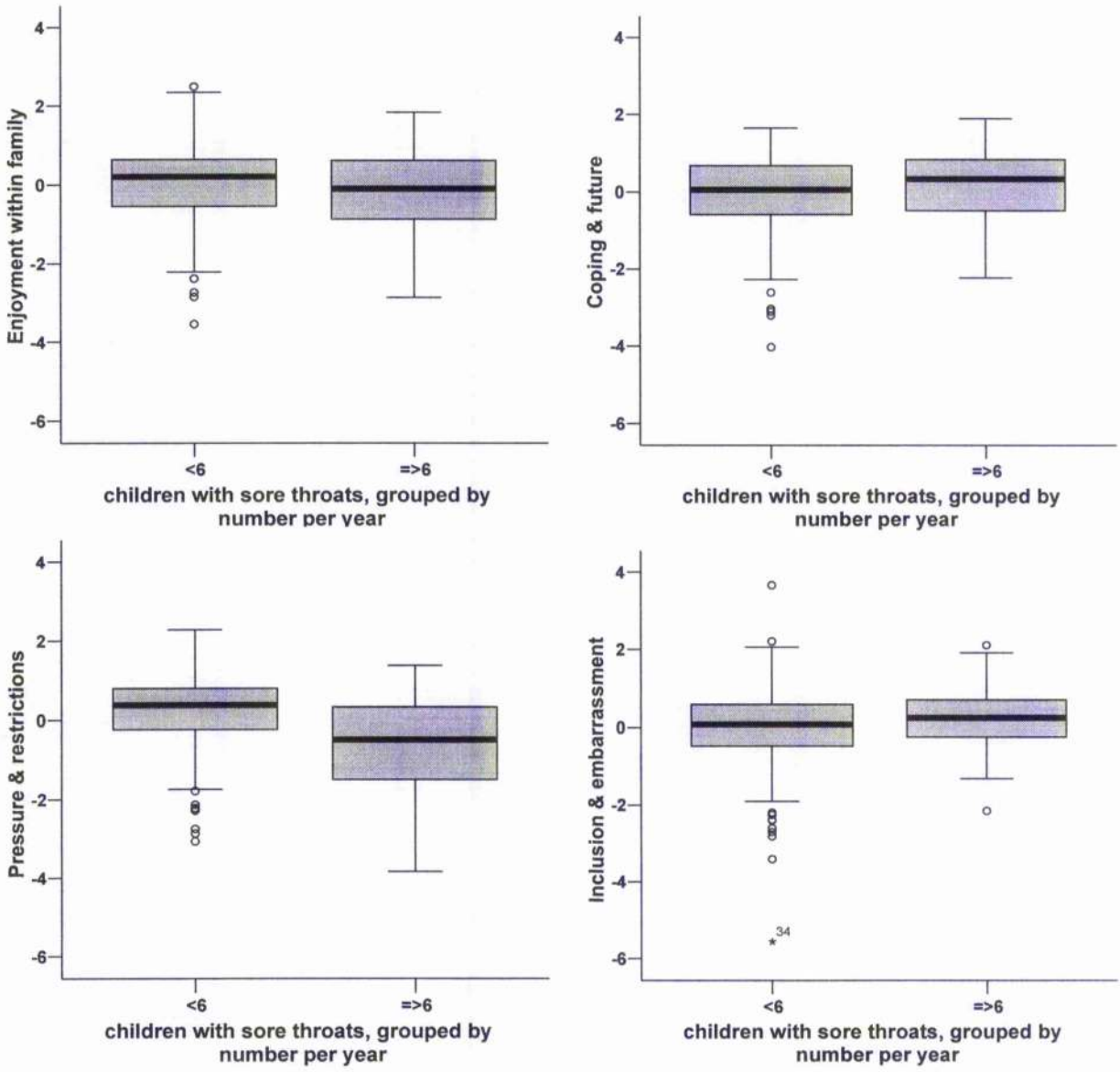


Table 6.1 Correlations between QOFL and CHQ domains

Table showing the Spearman correlation coefficients for 11 CHQ domains (excluding the 4 domains which consist of single-item responses) with the QOFL score. All correlations are significant, but the greatest correlation is for the family activities domain, as expected.

CHQ domain	**Spearman's rho**	**p**
Physical functioning	0.456	<0.001
Role/social limitations emotional	0.307	<0.001
Role/social limitations physical	0.310	<0.001
Bodily pain	0.210	0.019
Behaviour	0.495	<0.001
Mental health	0.482	<0.001
Self esteem	0.443	<0.001
General health perceptions	0.356	<0.001
Parental impact – emotion	0.447	<0.001
Parental impact – time	0.459	<0.001
Family activities	0.637	<0.001
Physical summary score	0.223	0.016
Psychosocial summary score	0.594	<0.001

Table 6.2 Item-total correlations for QOFL

Item-total correlations for the 26 items in the QOFL. The overall Cronbach's alpha was 0.9143. Alpha was only increased by the deletion of question 23.

QOFL item	Item-total correlation	Alpha if item deleted
q1. Enjoy going out together	0.5590	0.9104
q2. Restricted going out	0.4965	0.9115
q3. Effort getting ready	0.5620	0.9103
q4. Time for household activities	0.5168	0.9116
q5. Support	0.4102	0.9131
q6. Coping with life	0.6647	0.9084
q7. Future coping	0.7061	0.9077
q8. Enjoy TV together	0.4664	0.9120
q9. Time for leisure	0.6390	0.9088
q10. Enjoy meals	0.6257	0.9092
q11. Family agreement	0.5587	0.9104
q12. Outside interference	0.4148	0.9128
q13. Enjoy holiday	0.4791	0.9119
q14. Restrict holidays	0.3619	0.9138
q15. Pressure	0.5669	0.9102
q16. Worry when not together	0.4460	0.9139
q17. Satisfaction - achievement	0.5636	0.9103
q18. Stress of inclusion	0.5715	0.9102
q19. View of future	0.6749	0.9092
q20. Enough money	0.4586	0.9123
q21. Needs being met	0.6125	0.9097
q22. Embarassment at inclusion	0.3477	0.9137
q23. Understand condition	0.2175	0.9157 *
q24. Control over life	0.4237	0.9132
q25. Enjoy time at home	0.6438	0.9092
q26. Happiness	0.5874	0.9105

Table 6.3 QOFL factor loadings

Factor analysis of the 26 defined QOFL questions, using principle component extraction with varimax rotation. Only the first four factors with an eigenvalue greater than 1 were extracted. The table shows the factor loadings for each of the four factors, which have been given names for convenience. Only factor loadings of 0.3 or greater are shown for clarity.

Question	Factor			
	family enjoyment	coping & future	pressure & restrictions	inclusion & embarrassment
q1. Enjoy going out together	0.540		0.523	
q2. Restricted going out	0.438		0.608	
q3. Effort getting ready	0.441		0.389	
q4. Time for household activities	0.309	0.435		
q5. Support	0.440	0.367		
q6. Coping with life		0.636		
q7. Future coping	0.332	0.579	0.347	
q8. Enjoy TV together	0.533			
q9. Time for leisure	0.530		0.400	
q10. Enjoy meals	0.620			
q11. Family agreement	0.437	0.423		
q12. Outside interference			0.373	
q13. Enjoy holiday	0.620			
q14. Restrict holidays			0.604	
q15. Pressure		0.375	0.585	
q16. Worry when not together			0.674	
q17. Satisfaction – achievement	0.518			0.336
q18. Stress of inclusion	0.402		0.372	0.551
q19. View of future	0.428	0.608		
q20. Enough money		0.733		
q21. Needs being met		0.713		
q22. Embarassment at inclusion				0.545
q23. Understand condition				0.534
q24. Control over life		0.648		
q25. Enjoy time at home	0.685			0.351
q26. Happiness	0.452	0.474		0.419

7 The Glasgow Children's Benefit Inventory

7.1 Background

A measure of the benefit to the quality of a child's day to day life resulting from an intervention in children (such as surgery, hearing aid provision or advice in outpatients) would be very useful in clinical research. Although it is possible to measure change as the difference between two conventional health-related quality of life status instruments, one applied before the intervention, the other afterwards, a specifically-worded benefit measure would have a number of important advantages. It would be much more sensitive to change, free from the effects of reponse-shift bias and less prone to expectation bias [25]. In addition, such a measure can be retrospectively applied to a cohort of subjects who have undergone the intervention in the past, without the need for any questionnaires to be completed before the intervention.

A post-intervention health-related benefit measure, the Glasgow Benefit Inventory (GBI) [25], exists for use in adults and has been widely adopted for research in various aspects of otolaryngology, including tonsillectomy [28], snoring surgery [29], bone-anchored hearing aids [30], acoustic neuroma surgery [31], rhinoplasty [32] and speech therapy for dysphonia [33]. A benefit measure specific to surgery for obstructive sleep apnoea in children has also been reported, although not yet used widely [26].

7.2 Study aims

The aim of this study was to develop a generic health-related benefit measure appropriate for use in children and to assess aspects of its validity. The decision was made to develop a parent-completed instrument as the conditions of most interest in paediatric otolaryngology are most prevalent in pre-school age children who usually lack the necessary skills in language and abstract reasoning to complete such an instrument themselves. The proposed measure would be completed by parents on behalf of the child, and would be sufficiently broad in scope to be used for children of any age and in any area of paediatric medicine, not just otolaryngology.

7.3 Participants and methods

7.3.1 Question development

An initial list of potential items for inclusion was generated by

1. Studying existing well-known generic children's health-related quality of life instruments, namely the Child Health Questionnaire [56], The TACQOL [69] and TAPQOL [70] questionnaires, and the Health Utilities Index mark II and mark III [18].

2. Reviewing the published literature on health-related quality of life assessment in children.
3. Drawing from the experience gained in developing the GBI [25] to identify areas applicable to adults that might be generalised to children.
4. Semi-structured interviews with the parents of children who had previously undergone a range of otolaryngological operations, to determine which areas of the child's life had been changed (for better or worse) by the surgery (Table 7.1).

The resulting items were used to compose a draft Glasgow Children's Benefit Inventory (GCBI), consisting of 24 questions (see appendix). The format for the questions was based on that used for the GBI [25]. Each question is worded with reference to the time since a specified intervention. This could be worded to refer to any intervention, such as hearing aid provision, but for the purpose of this study it was worded with reference to the child's operation. For each question, a response is given on a five-point Likert-type rating scale, with the central point being "no change" and the extremes representing "much better" and "much worse". The order of the response scale was initially varied randomly with a positive response being on the right side approximately half the time, and on the left side for the remainder. This was done to control for a form of response bias.

The draft questionnaire was then piloted on a group of 11 parents to obtain their comments on clarity, ease of use, and relevance, with suggestions for

improvement. As a result, a number of minor changes to the wording were made. The decision was also made to have all the responses ordered in the same direction, rather than randomly varied, as the parents found this confusing and unhelpful (a similar sequence of events occurred in the design of the Abbreviated Profile of Hearing Aid Benefit [116]).

7.3.2 *Instrument validation*

The revised questionnaire was then sent out by post with a covering letter and postage-paid return envelope to the parents of all children who had undergone tonsillectomy or ventilation tube insertion between January 1998 and December 2001 at Crosshouse Hospital, Kilmarnock. No financial incentives for returning the questionnaire were offered. Prior approval was obtained from the hospital's research ethics committee.

Tonsillectomy and ventilation tube insertion were chosen for study because they are the most commonly performed surgical procedures in children in the UK. In addition, they are not life-saving procedures, but are performed with the intention of improving the child's quality of life, and parental satisfaction in this regard is very high [62].

When questionnaires are sent out unexpectedly some years after hospital treatment it is inevitable that the response rate will be low. Based on experience

from previous studies, we estimated that no more than 45% [25], and perhaps as few as 25%, of questionnaires would be completed and returned.

Our aim was to establish the convergent validity of the GCBI by showing that the results correlate with a measure of the technical success of the procedure. By technical success, we mean some potentially quantifiable assessment that the operation has produced the desired medical outcome which, in turn, we expect to influence the child's quality of life. We, therefore, included a short clinical questionnaire with the mailshot. This included a question about the parent's overall satisfaction with the surgery, on a five-point Likert rating scale from "very unhappy" to "very happy". It also included questions on the child's improvement in hearing and speech, and the frequency of sore throat or ear infections (as relevant) since the operation.

It was a condition of ethical committee approval that the study be entirely anonymised, so that no linkage of results with hospital records or clinical findings was possible. The use of audiometric outcomes was therefore impossible. To address this, we included the MRC's 4-item Reported Hearing Disability Scale (RHD-4), which is a validated measure of hearing disability as reported by the parents, and based on questions from the MRC multi-centre TARGET Trial (Mark Haggard, MRC Institute of Hearing Research, personal communication). Thus, we intended to compare the results of the GCBI with various measures of technical success, comprising frequency of sore throats or ear infections, reported hearing disability, and overall parental satisfaction with

surgery. A similar methodology was used successfully in the validation of the GBI [25].

The list of children was obtained from the hospital's computerised theatre records. Every entry on the computerised list was checked individually to remove duplicate entries and to ensure that only correct procedures were included. Where ventilation tube insertion was performed as a prelude to evoked response audiometry in the investigation of possible sensorineural hearing impairment, the child was removed from the study list. Similarly, where the child had undergone both tonsillectomy and ventilation tube insertion at the same time, the child was not included in the study. Minor nasal procedures (such as adenoidectomy, antral lavage, or submucous diathermy to the turbinates) performed at the same time as the tonsillectomy or ventilation tube insertion were not judged to be of significance for the purpose of the study, and these children were not excluded.

Data were stored on computer and analysed using SPSS version 11.0.

7.4 Results

The GCBI was sent out to the parents of 1777 children, comprising 924 girls and 853 boys. They ranged in age from 1 to 15 years (median 6, mean 7.18) at the time of surgery. 1234 had undergone tonsillectomy (without ventilation tube

insertion, but some with minor nasal procedures) and 543 had undergone ventilation tube insertion (unilateral or bilateral, without tonsillectomy, but some with minor nasal procedures).

Completed questionnaires were returned for 670 children (38%), of whom 452 had undergone tonsillectomy and 218 had undergone ventilation tube insertion. The 670 questionnaires contained a total of 16,080 items, of which only 93 (0.6%) were left uncompleted.

A summary score for the GCBI was calculated by assigning the individual question responses a numerical value from -2 to +2, then adding these up, dividing by the number of questions (24) and multiplying by 50 to produce a result on a scale from -100 (maximum harm) to +100 (maximum benefit). If 5 or fewer questions had missing values, the missing values were imputed using the overall average for that questionnaire. We had planned to regard questionaires with more than 5 missing values as unsuitable for analysis, but there were no such questionnaires in this study. The results in this study population were widely spread between -44 and +100 with a median of 29, a mean of 33 and a standard deviation of 24 (Figure 7.1).

In the children who had undergone tonsillectomy, 173 were reported as having had no sore throats since the operation, 231 had had some sore throats but not as many as before the surgery, and 13 were having as many sore throats as before. The median scores in these groups were 35, 31 and 0 respectively

(Kruskal Wallis, $p<0.001$ for a significant difference, largely due to the difference between the third group and the other two; Figure 7.2). The parents rated their overall satisfaction with surgery on a 5 point Likert scale: 340 reported themselves "very happy", 87 "happy", 21 "not sure", 3 "unhappy" and none "very unhappy". The median scores in these groups were 38, 21, 0, and 0 respectively (Jonckheere Terpstra test, $p<0.001$; Figure 7.3).

In the children who had undergone ventilation tube insertion, and excluding those who said the child had never had ear infections before surgery, 73 were reported as having had no ear infections since the operation, 97 had had some ear infections but not as many as before the surgery, and 13 were having as many ear infections as before. The median scores in these groups were 35, 25 and 0 respectively (Kruskal Wallis, $p<0.001$, largely due to the difference between the third group and the other two; Figure 7.4).

In the children who had undergone ventilation tube insertion, and excluding those who said the child had never had any hearing or speech concerns before surgery, 162 were reported as being much improved and 30 as having had no improvement. The median scores in these groups were 29 and 3 respectively (Mann Whitney, $p<0.001$; Figure 7.5). There was some weak correlation between the GCBI scores and the degree of residual hearing disability as assessed by RHD-4 (Spearman's rho=-0.193, $p=0.005$, $n=214$; Figure 7.6).

Overall satisfaction with surgery for all the children who had undergone ventilation tube insertion was as follows: 124 reported themselves "very happy", 58 "happy", 31 "not sure", 4 "unhappy" and none "very unhappy". The median scores in these groups were 35, 19, 4 and 0 respectively (Jonckheere Terpstra, $p<0.001$; Figure 7.7). For the whole group of children in the study (tonsillectomy and ventilation tubes combined) the levels of reported satisfaction were 464 "very happy", 145 "happy", 52 "not sure", 7 "unhappy" and none "very unhappy", with median values of 35, 19, 2 and 0 respectively (Jonckheere Terpstra, $p<0.001$; Figure 7.8).

The GCBI questionnaire had a high level of internal consistency (Cronbach's alpha=0.92). All the individual items were positively correlated with the total score, such that alpha was never increased if an item was deleted (Table 7.2).

Factor analysis was performed by principle component extraction with varimax rotation, selecting for factors with eigenvalues greater than 1. Four factors were extracted, which between them accounted for 62% of the variance. The extent to which each item in the GCBI loaded onto each of these four factors is shown in Table 7.3.

The first factor was most heavily loaded onto by the questions relating to self-consciousness, family harmony, embarrassment, easy distraction, self-esteem, confidence and self-care: for convenience, we have chosen to label this factor "emotion". The second factor was most heavily loaded onto by the questions

relating to overall life, time off school, colds, visits to the doctor and need for medication: we have labelled this factor "physical health". The third factor was most heavily loaded onto by the questions about progress and development, easy distraction, learning and concentration: we have labelled this "learning". The fourth factor was most heavily loaded onto by the questions about liveliness, sleep, food, fun with friends and leisure: we have labelled this "vitality".

The factor scores did not differ significantly between the children who had undergone tonsillectomy and those who had undergone ventilation tube insertion. Within the tonsillectomy group, the "physical health" and "vitality" factor scores were associated with the number of sore throats since surgery (One-way ANOVA, $p<0.001$ and $p=0.031$ respectively – Figure 7.9). Three of the four factor scores were associated with parental satisfaction (One-way ANOVA with linear trend; "physical health" $p<0.001$, "learning" $p=0.011$, "vitality" $p<0.001$; Figure 7.10).

Within the ventilation tube group, the "physical health" and "learning" factor scores were associated with hearing and speech improvement (t-test, $p<0.001$ in both cases – Figure 7.11). The "physical health" and "emotions" factor scores were associated with number of ear infections since surgery (One-way ANOVA with linear trend; "learning" $p<0.001$, "emotions" $p=0.046$; Figure 7.12). The "learning" and "physical health" factor scores were associated with overall

parental satisfaction with surgery (One-way ANOVA with linear trend, $p<0.001$ in both cases – Figure 7.13).

7.5 Discussion

This retrospective approach to measuring benefit from an intervention such as surgery suffers from a number of drawbacks, most notably bias related to the parents' prior expectation of benefit, and changes in the perceived effect of the intervention with increasing time. These criticisms can also be applied, however, to the measurement of benefit as the difference in a measure applied before and after the intervention. The great advantages of the retrospective approach are that it halves the burden of questionnaires for the parents (thereby increasing compliance), it is much more sensitive to change and it can be used in rarer conditions where a sizeable cohort of patients can take years to build up. An appropriate benefit measure could, therefore, have widespread application in paediatric medicine, and paediatric otolaryngology in particular. Certainly, the analogous adult instrument, the GBI, has been found to be useful in the assessment of outcomes in a variety of circumstances [29 32 28 30 31 33].

Our intention was to create a generic instrument which would be applicable regardless of the child's age, and considerable effort was put into the phrasing of the questions with this aim in mind. Parents were involved at all stages to ensure that the instrument addressed the issues of importance to them, rather

than only addressing the concerns of clinicians. In addition, we wanted to create an instrument that addressed social, psychological and physical functioning and the effect of disease on day to day activities, without reference to any specific symptoms or diseases, so that the instrument could be as widely applicable as possible.

We performed this study with the intention of demonstrating aspects of reliability and validity in the context of commonly performed otolaryngological surgery. Reliability, in the sense of freedom from random error in the measurement, is shown by a high degree of internal consistency and by the presence of a coherent and clearly interpretable factor structure. Such a high degree of internal consistency could be considered as an indication that there is redundancy in the questions, and that a much shorter version of the instrument could be produced. While this may be true, the wide range of questions used allows us to produce much richer data for factor analysis. The factor scores may prove to be more informative than a simple summary score. The factor scores vary in a way that one might expect, with "physical health" varying with frequency of sore throats and ear infections, and "learning" varying with reported benefit to hearing and speech.

Validity depends on context, but we have been able to demonstrate that in this study population the scores obtained with the GCBI behave in a predictable and logical way when compared against measures of the technical success (residual ear and throat infections after surgery, subjective improvement in hearing and

speech) and overall level of parental satisfaction with surgery. Also, the outcome scale from –100 to +100 has been shown to be anchored appropriately, with groups of children with no reported benefit from surgery having median GCBI scores of zero. Ideally, we would have liked to compare the GCBI scores against more objective outcome measures such as pure tone audiometry, but the terms of ethical approval for the study prevented linkage of the questionnaires with clinical records.

In a study of this type, multiple statistical comparisons are unavoidable, and values for statistical significance should be interpreted with caution as a result. This is also true for correlations which were of themselves weak, but highly statistically significant nonetheless due to the large number of subjects studied.

In summary, the GCBI is a means to retrospectively assess benefit after an intervention in children and we have shown initial evidence of reliability and validity. Although not restricted to any branch of paediatric medicine, it is eminently suitable for use in paediatric otolaryngology.

Figure 7.1 Range of GCBI scores

Histogram to show the distribution of GCBI scores in the study population. Each bar represents the total for scores over a ten-point range, from the worst possible (-100, maximum harm) to the best possible (+100, maximum benefit).

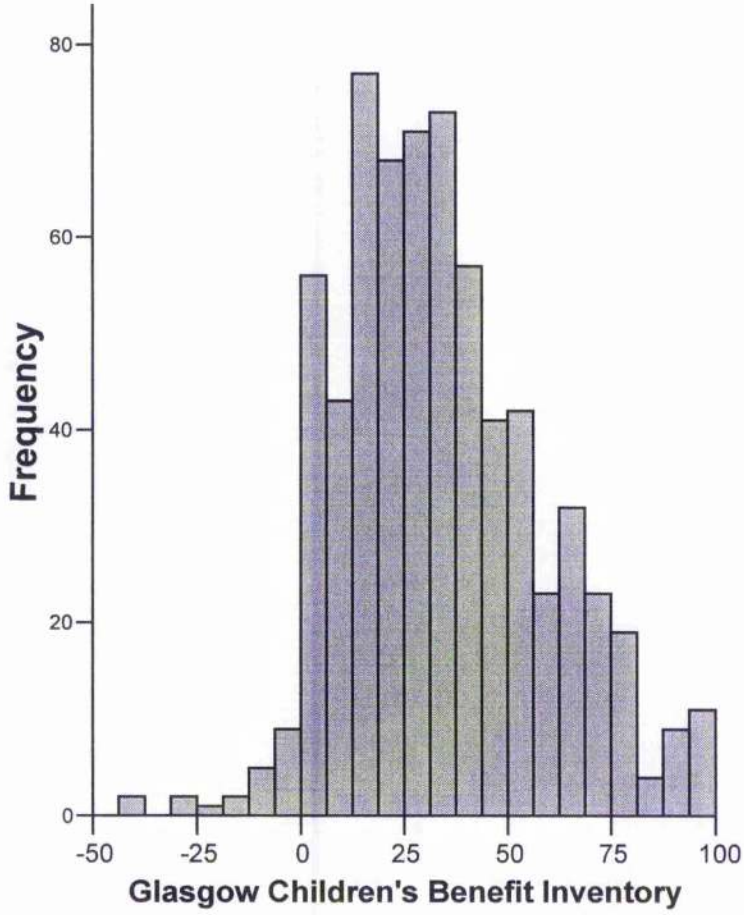


Figure 7.2 GCBI scores and outcome of tonsillectomy

Boxplot of GCBI scores for children who have undergone tonsillectomy, grouped according to the number of sore throats suffered since surgery, as a measure of the technical success of the surgery.

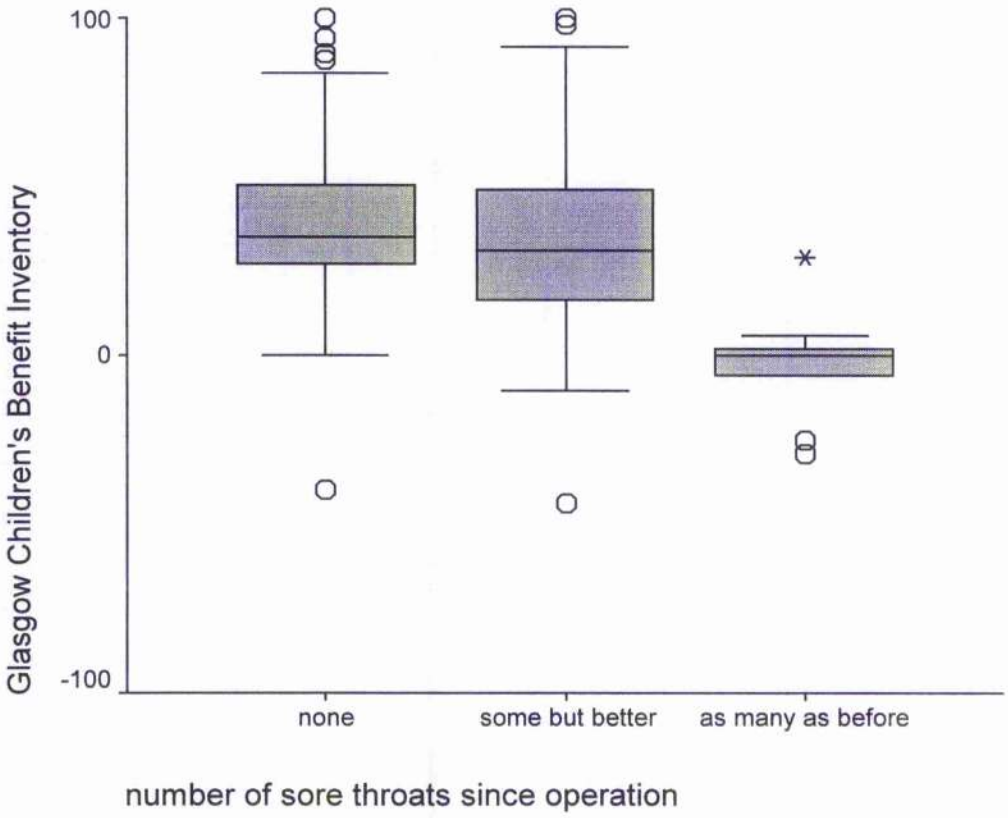


Figure 7.3 GCBI scores and parental satisfaction (tonsillectomy)

Boxplot of GCBI scores for children who have undergone tonsillectomy, grouped by the degree of parental satisfaction with surgery as rated on a 5-point Likert scale from "very happy" to "very unhappy" (none chose "very unhappy").

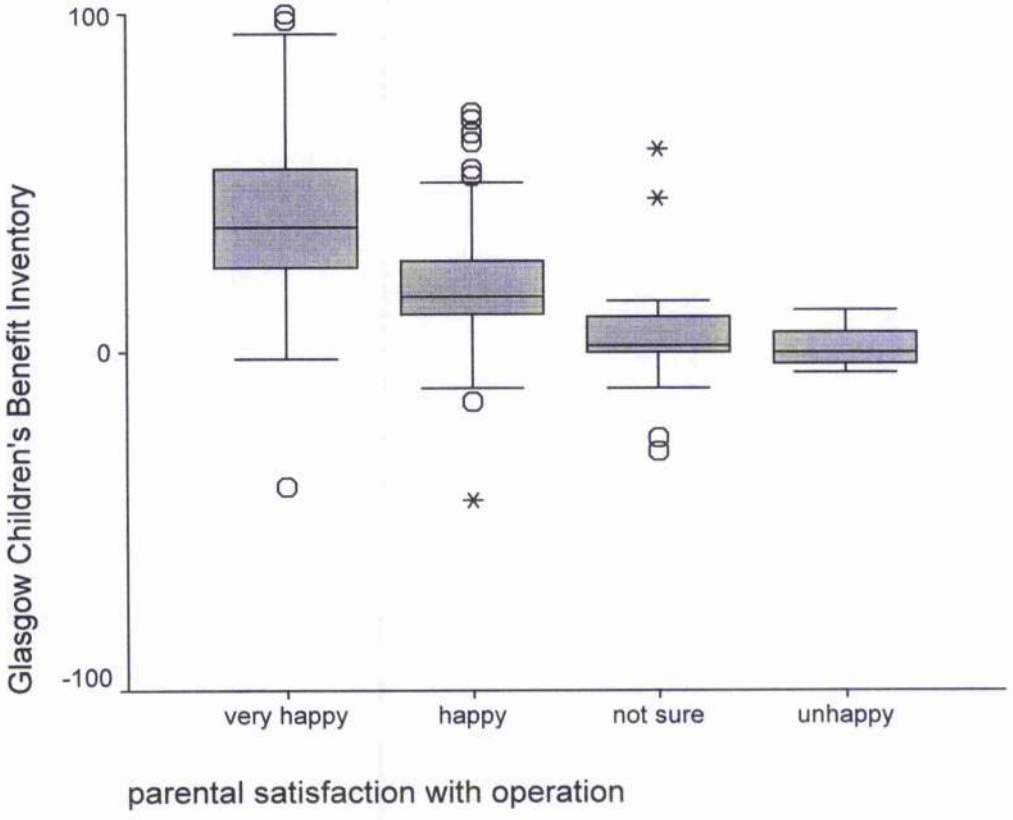


Figure 7.4 GCBI scores and outcome of ventilation tubes (infections)

Boxplot of GCBI sores for children with a history of ear infections who underwent ventilation tube insertion, grouped according to the number of ear infections suffered since surgery, as a measure of the technical success of the surgery.

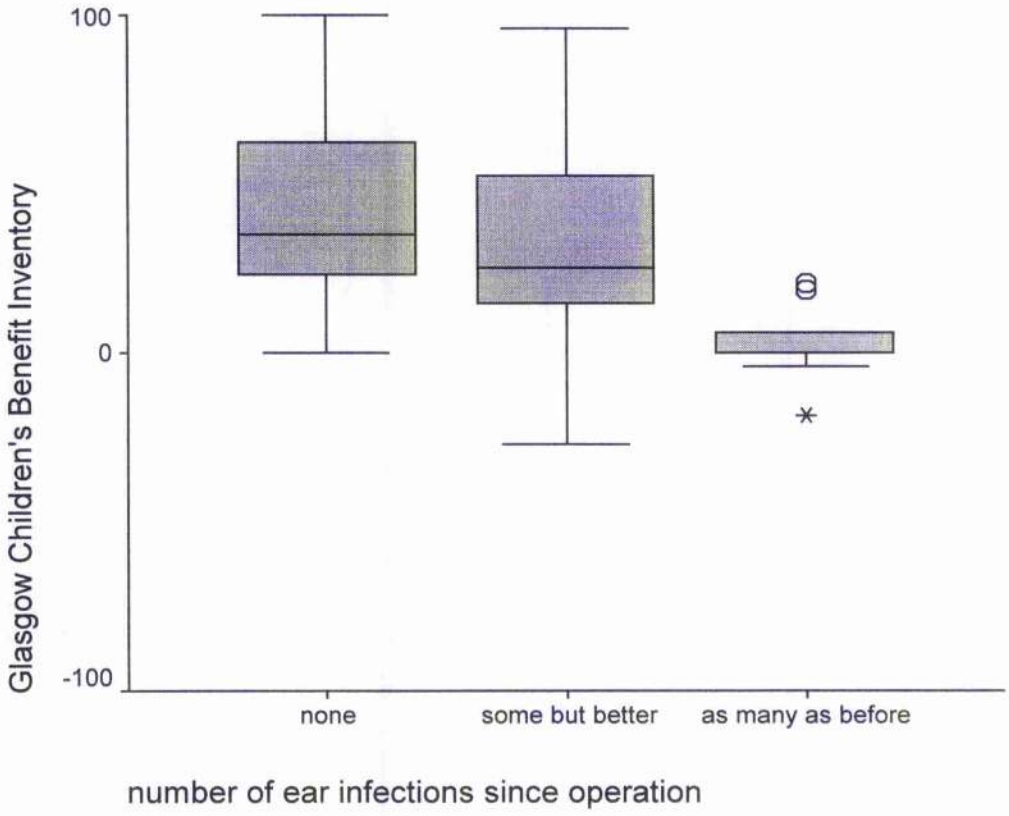


Figure 7.5 GCBI scores and outcome of ventilation tubes (hearing)

Boxplot of GCBI scores for children who have undergone ventilation tube insertion and where there were concerns about hearing or speech pre-operatively, grouped according to the parents' rating of the effect of surgery on the hearing and speech.

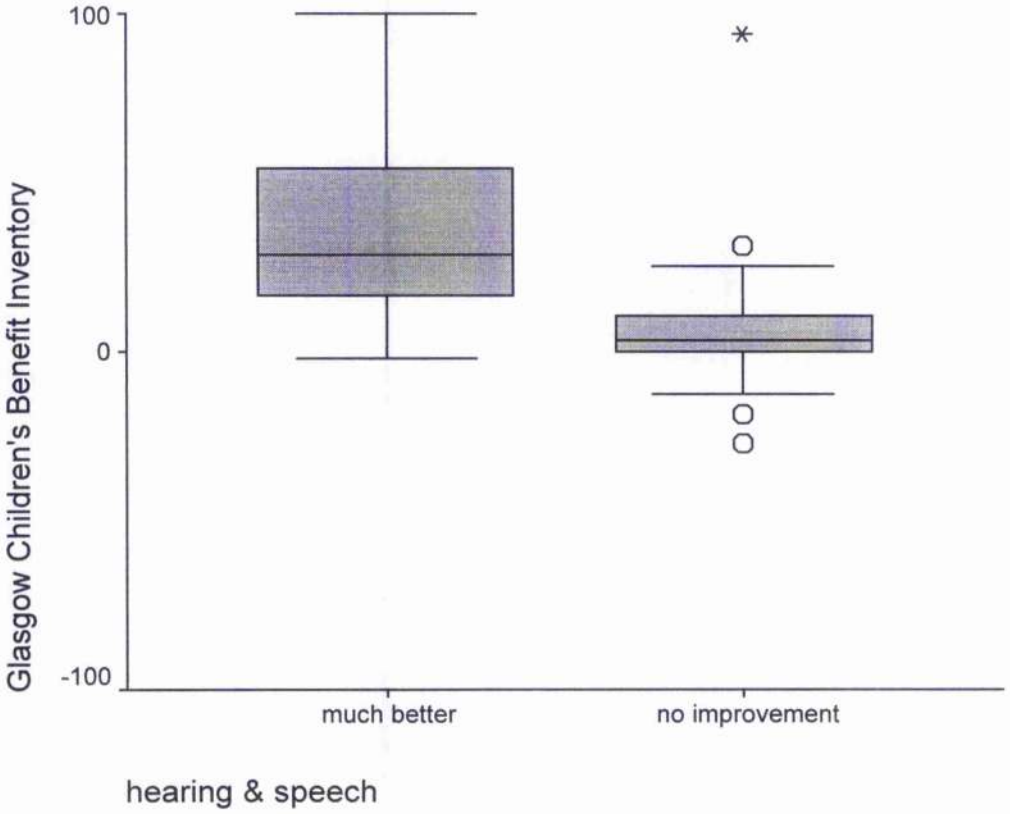


Figure 7.6 GCBI scores and RHD-4

Scatterplot of GCBI score against the child's residual hearing disability as rated using RHD-4 (higher RHD-4 scores reflect greater hearing concern).

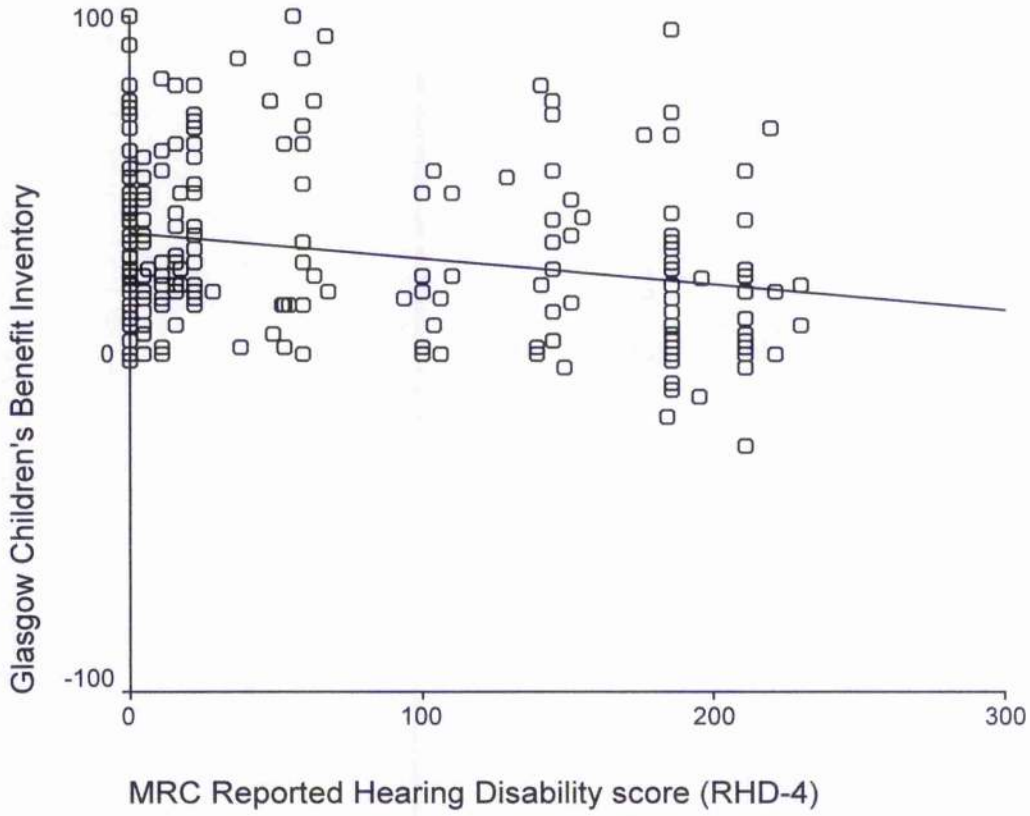


Figure 7.7 GCBI scores and parental satisfaction (ventilation tubes)

Boxplot of GCBI scores for the children who underwent ventilation tube insertion, grouped by the degree of parental satisfaction with surgery as rated on a 5-point Likert scale from "very happy" to "very unhappy" (none chose "very unhappy").

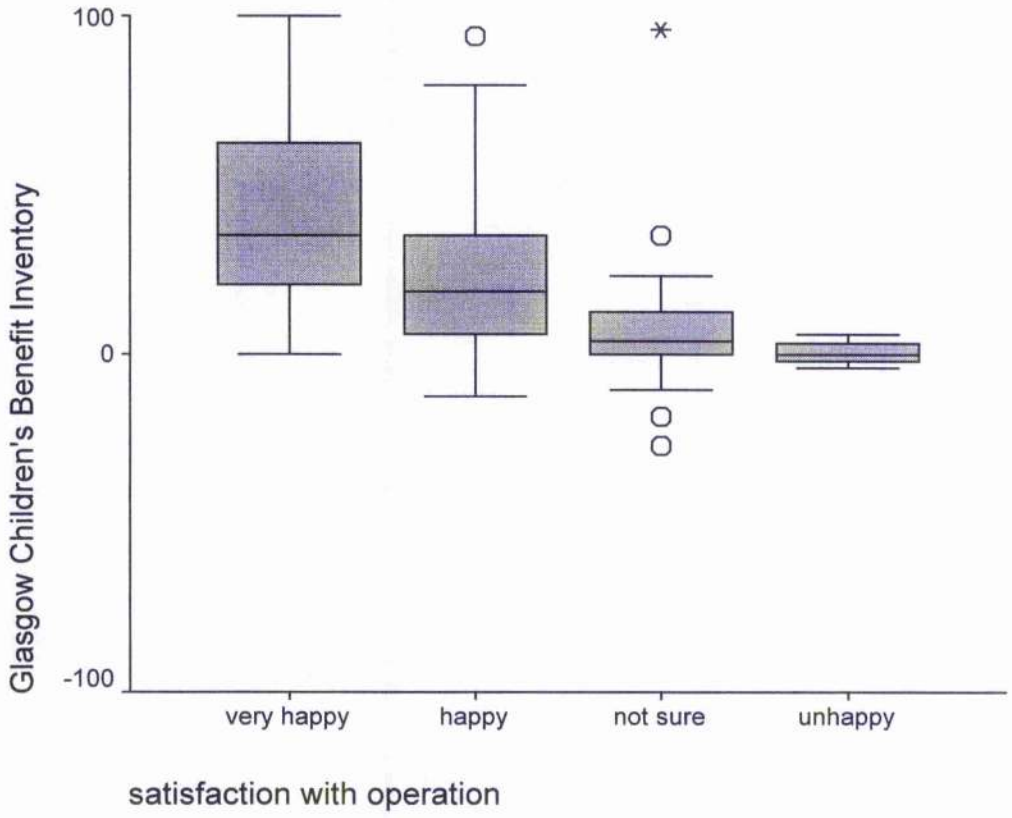


Figure 7.8 GCBI scores and parental satisfaction (overall)

Boxplot of GCBI scores for all the children in the study (tonsillectomy and ventilation tube insertion), grouped by the degree of parental satisfaction with surgery as rated on a 5-point Likert scale from "very happy" to "very unhappy" (none chose "very unhappy").

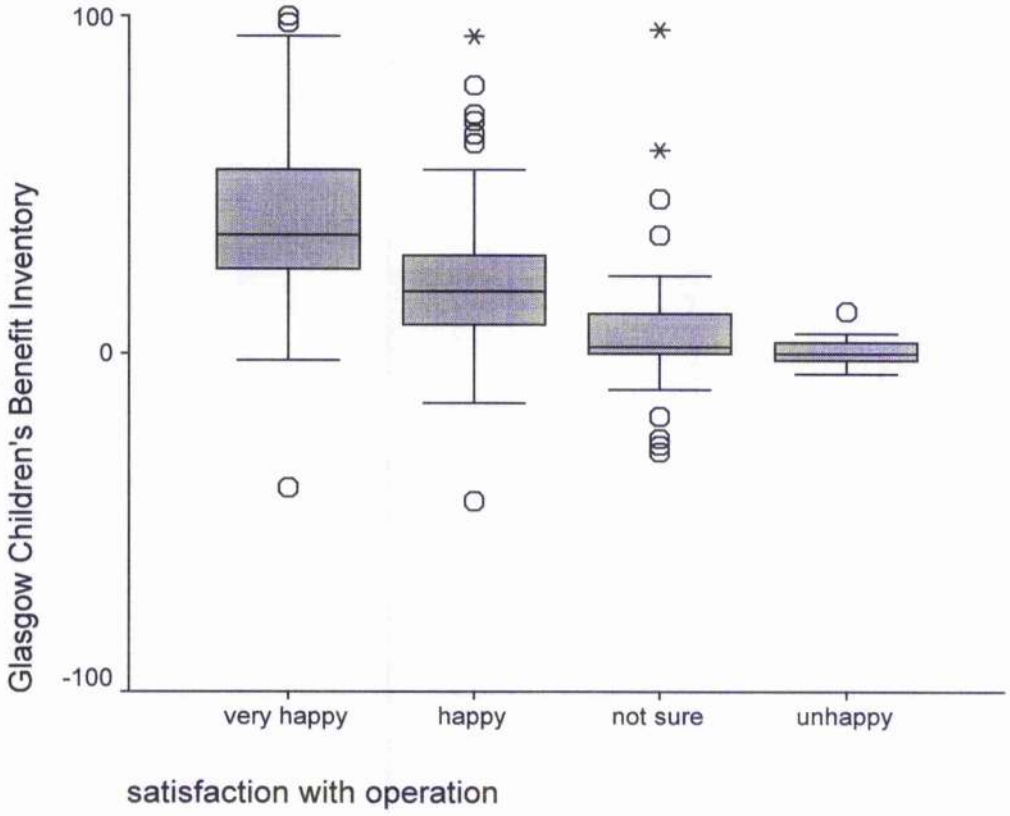


Figure 7.9 GCBI factor scores and outcome of tonsillectomy

Four boxplots, showing how the four factor scores in the GCBI (emotion, physical health, learning and vitality) relate to the frequency of residual sore throats after surgery in the subgroup of children who underwent tonsillectomy (n=372).

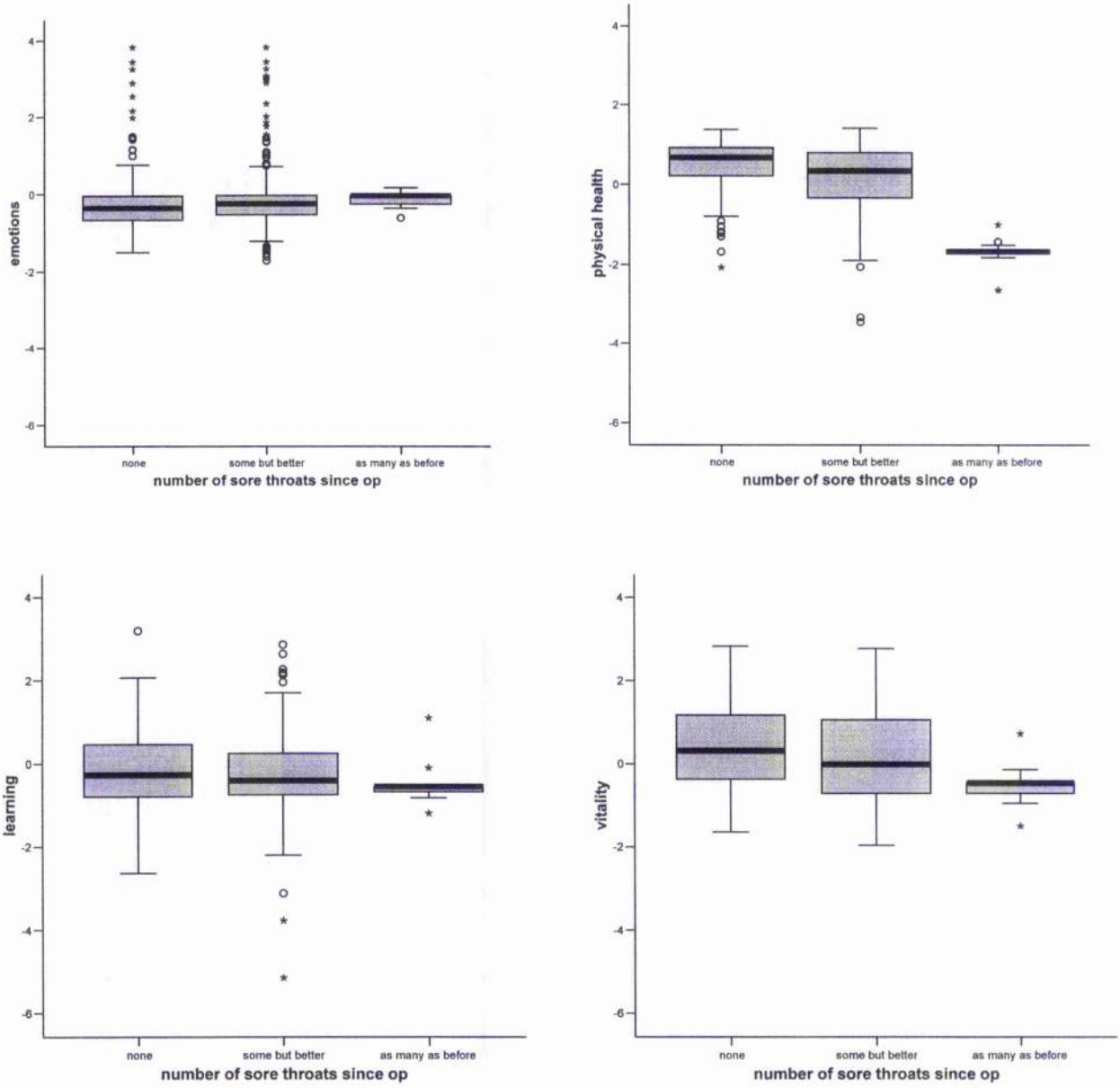


Figure 7.10 GCBI factor scores and satisfaction with tonsillectomy

Four boxplots, showing how the four factor scores in the GCBI (emotion, physical health, learning and vitality) relate to overall parental satisfaction with surgery in the subgroup of children who underwent tonsillectomy (n=372).

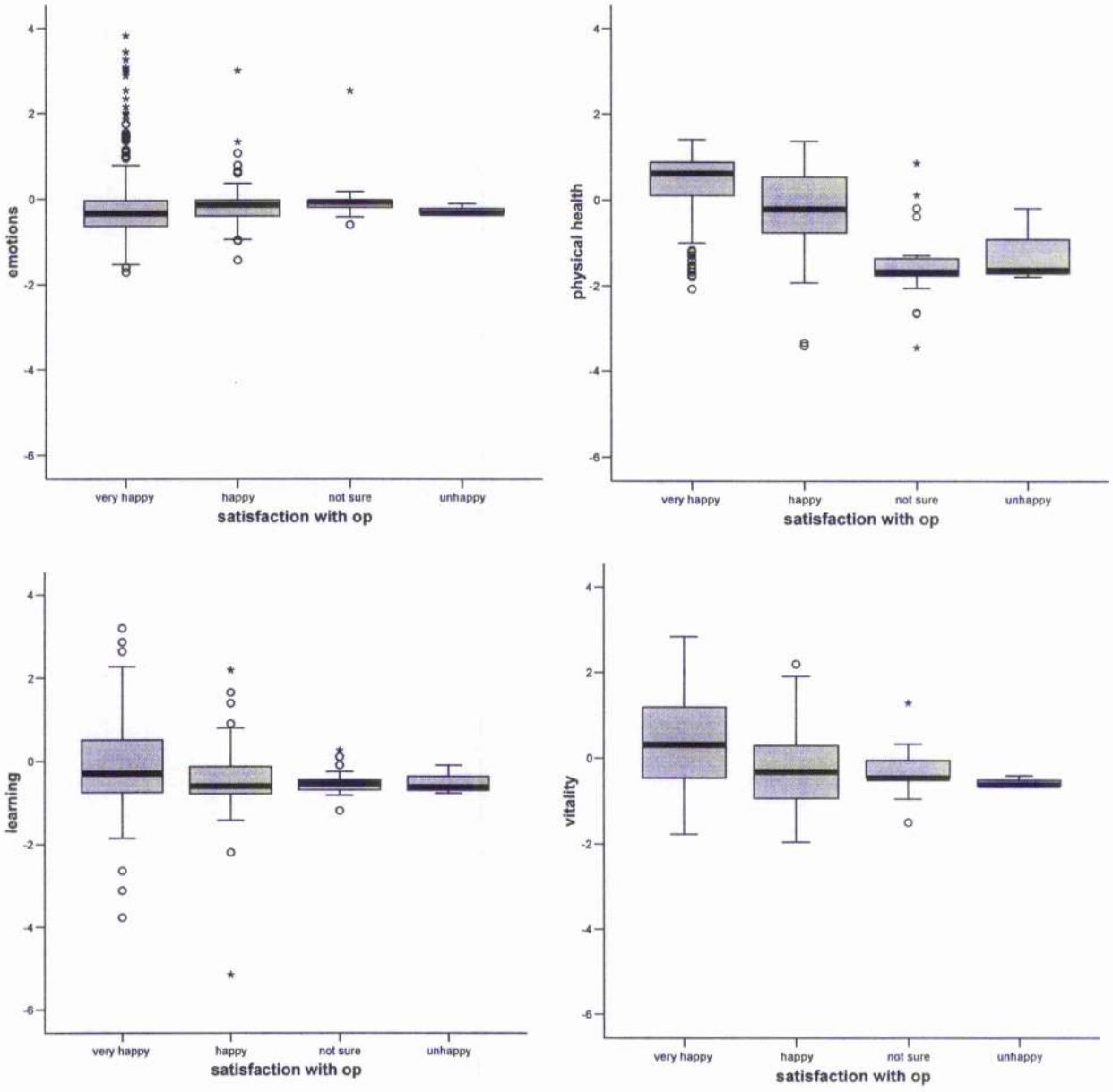


Figure 7.11 GCBI factor scores and hearing

Four boxplots, showing how the four factor scores in the GCBI (emotion, physical health, learning and vitality) relate to reported improvement in hearing and speech after surgery in the subgroup of children who underwent ventilation tube insertion (n=177).

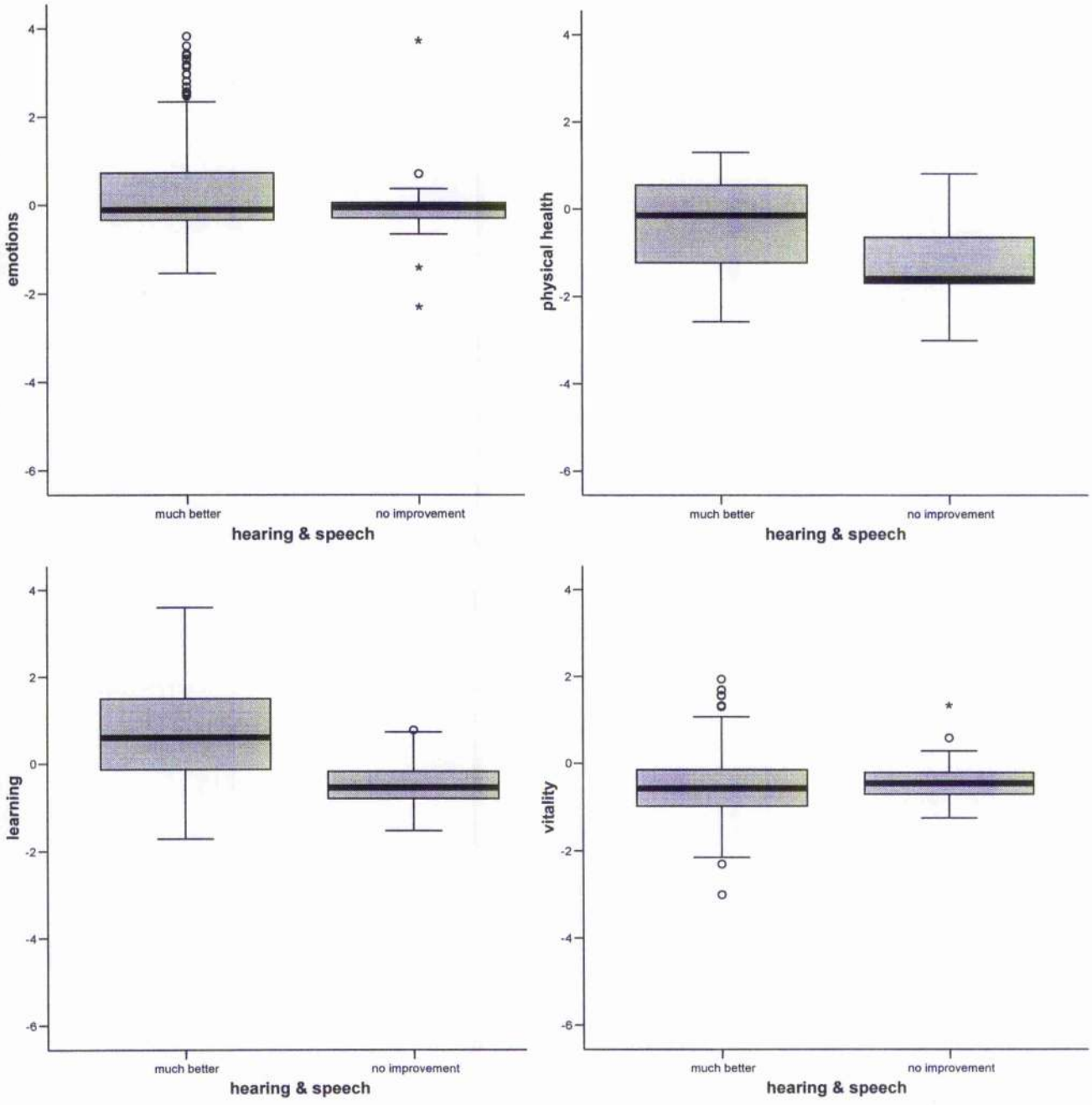


Figure 7.12 GCBI factor scores and ear infections

Four boxplots, showing how the four factor scores in the GCBI (emotion, physical health, learning and vitality) relate to frequency of residual ear infections after surgery in the subgroup of children who underwent ventilation tube insertion (n=168).

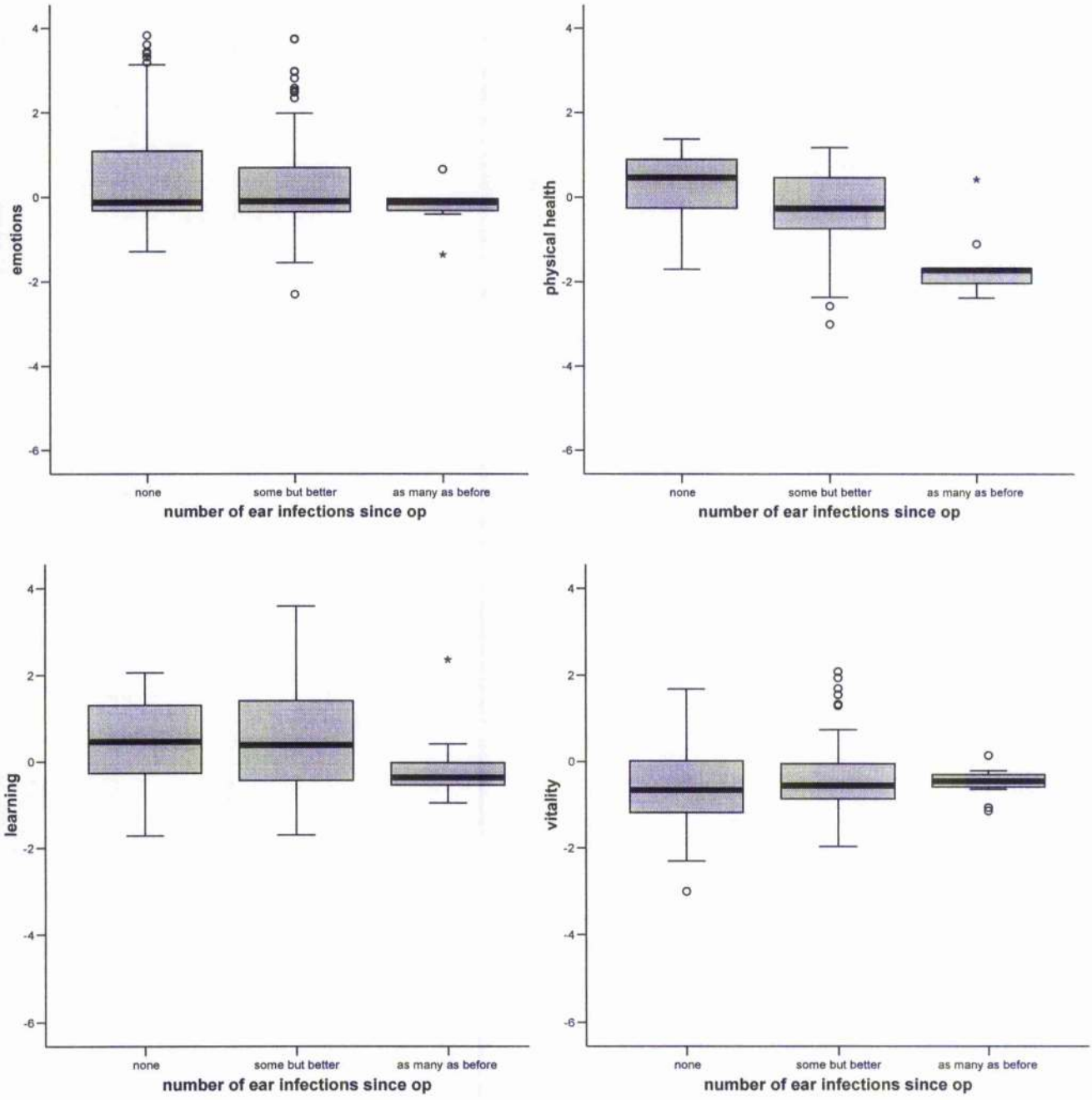


Figure 7.13 GCBI factor scores and satisfaction with ventilation tubes

Four boxplots, showing how the four factor scores in the GCBI (emotion, physical health, learning and vitality) relate to overall parental satisfaction with surgery in the subgroup of children who underwent ventilation tube insertion (n=202).

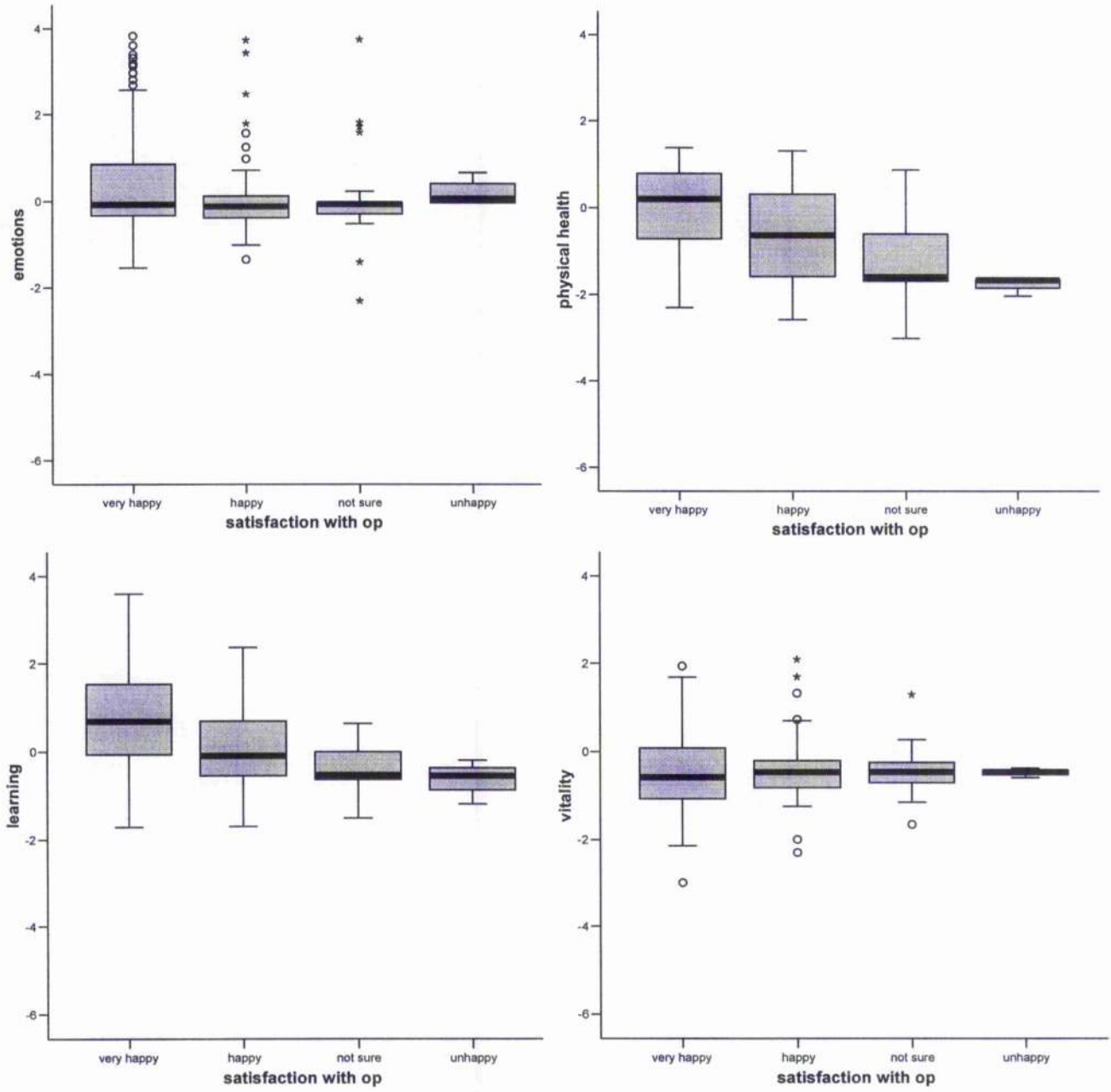


Table 7.1 Aspects of life improved by otolaryngological surgery

In semi-structured interviews, the parents of 6 children aged 3-13 years who had undergone routine otolaryngological surgical procedures reported the following areas in which life had been improved by the surgery.

Physical symptoms
Sore throats
Snoring
Hearing
Speech
Blocked nose and catarrh
Pain
General health
Quality of sleep
Temper, irritability
Mood
"Clinginess"
Time off nursery/school
Participation in sports
Attention
Confidence
Eating/appetite
Parents/Family
Sleep
Time off work
Communication with child
Disturbance due to child's noisey breathing
Relationship with child
Visits to doctor
Need for TV to be loud

Table 7.2 GCBI item-total correlations

Item-total correlations for the items in the GCBI. Cronbach's alpha overall is 0.9302. The right-hand column shows the extent to which alpha is changed if that particular item is deleted (alpha is never increased by deleting an item, showing that they are all positively correlated).

GCBI Item	Item-total correlation	Alpha if item deleted
q1. Overall life	0.5900	0.9271
q2. Things they do	0.6320	0.9264
q3. Behaviour	0.5645	0.9275
q4. Progress & development	0.6223	0.9266
q5. Liveliness	0.6654	0.9258
q6. Sleep	0.5716	0.9276
q7. Food	0.4902	0.9289
q8. Self consciousness	0.5914	0.9275
q9. Family harmony	0.5660	0.9277
q10. Fun with friends	0.6722	0.9257
q11. Embarrassment	0.4956	0.9287
q12. Easily distracted	0.5191	0.9282
q13. Learning	0.5778	0.9273
q14. Time off school	0.5348	0.9281
q15. Concentration	0.5804	0.9273
q16. Irritability	0.6697	0.9258
q17. Self-esteem	0.6440	0.9263
q18. Happiness	0.6851	0.9255
q19. Confidence	0.6072	0.9270
q20. Self-care	0.4308	0.9295
q21. Leisure	0.6010	0.9269
q22. Catches colds	0.4942	0.9291
q23. Visits to doctor	0.5691	0.9275
q24. Need for medication	0.5680	0.9276

Table 7.3 GCBI factor loadings

Factor analysis of the 24 GCBI questions, using principle component extraction with varimax rotation. Factors with an eigenvalue greater than 1 were extracted. The table shows the factor loadings for each of the four factors, which have been given names for convenience. Only factor loadings of 0.3 or greater are shown for clarity.

Question	Factor			
	emotion	physical health	learning	vitality
q1. Overall life		0.673	0.303	
q2. Things they do			0.496	0.433
q3. Behaviour	0.459		0.453	
q4. Progress & development			0.757	
q5. Liveliness				0.729
q6. Sleep		0.301		0.671
q7. Food				0.705
q8. Self consciousness	0.782			
q9. Family harmony	0.682		0.352	
q10. Fun with friends	0.337			0.593
q11. Embarrassment	0.855			
q12. Easily distracted	0.512		0.547	
q13. Learning			0.775	
q14. Time off school		0.718		0.306
q15. Concentration	0.404		0.631	
q16. Irritability	0.387		0.467	0.346
q17. Self-esteem	0.671		0.309	
q18. Happiness	0.399		0.346	0.460
q19. Confidence	0.689		0.359	
q20. Self-care	0.531			0.380
q21. Leisure		0.320		0.600
q22. Catches colds		0.802		
q23. Visits to doctor		0.901		
q24. Need for medication		0.882		

8 Conclusions

Otolaryngologists operate on more children than any other surgical specialty in the UK [3]. Most of these interventions are aimed at relieving symptoms rather than prolonging lives. The assessment of outcomes is best done, therefore, using some sort of measures of quality of life, but we must be careful that our choice of measuring instrument is appropriate and justifiable. The range of measures available may seem bewildering and choosing the most appropriate for a particular situation can appear complex.

Many instruments are reported as being generic, meaning that they are felt to be applicable to any disease. None could ever have been tested in every disease, and the statement that an instrument is generic is largely an opinion. Many instruments are designed by people experienced in general paediatric medicine, so they will be designed with certain diseases in mind – asthma, chronic arthritis, skin conditions and so on. It cannot be assumed that they will perform well in an otolaryngology setting. In this thesis, a range of generic instruments have been evaluated in the context of three common conditions in paediatric otolaryngology: OME, recurrent AOM and sore throats. The aim was to compare the discriminative ability of these instruments in children with varying severities of disease.

The instruments were chosen as the most likely to perform well in this setting, although none was felt initially to be ideal. TAPQOL is the only one designed

for the very young children (aged 1-5 years) so common in otolaryngology clinics. TACQOL is the only one to include a question on sore throats and ear infections. Both TACQOL and TAPQOL are cumbersome to use, however, as there is no summary score, just a large number of domain scores. In addition, although they are presented as being complementary to each other, the two instruments are far from comparable in the range of domains, or in the way the scores are reported. TACQOL is only suitable for a minority of otolaryngology patients because of its age range (6+). The CHQ is probably the most well-known and widely-used of the available instruments and is becoming close to a "gold standard". It is also rather cumbersome to use, however, as the calculation of scores is extremely complex. It also contains few questions that relate to ear, nose and throat issues, and communication issues in particular. The HUI mark III is a health utility measure which can be used for economic evaluations, and it has specific questions on hearing and speech. It fails to address social or psychological issues to any useful extent, however.

The results of the studies presented above show that, despite the potential drawbacks of each of these instruments, all performed well in the otolayngology setting. Each instrument was shown to be free from significant influence of extraneous variables such as age, sex and socio-economic deprivation. Indeed, the HUI mark III could possibly be used beyond its suggested lower age limit of 6 years, perhaps in children as young as 4, thus increasing its usefulness in otolaryngology. All the instruments showed some degree of association between their scores for HRQOL and the severity of the

underlying disease, in the manner that one would expect. There is some evidence, however, that both the CHQ and TAPQOL lack some sensitivity to the impairments present in children with OME. This reflects the concerns mentioned above regarding the fact that so-called generic instruments often fail completely to address communication issues, so important in otolaryngology.

It was consistently found that recurrent AOM was associated with poorer HRQOL ratings than OME or sore throats. While this may simply reflect the lack of sensitivity to the impairments present in OME, it was such a consistent finding with different instruments that it may will be genuine. Anecdotally, the pain, fever and sleepless nights that ear infections cause do seem to be perceived by parents as a very significant burden of ill health.

In fact, the scores for HRQOL reported here with the various instruments do make for interesting comparisons with published values for children with other health complaints. Certainly, OME, recurrent AOM and sore throats are all associated with scores for HRQOL that are considerably worse than for healthy children, and in many cases as bad or worse than scores for chronic conditions such as asthma, juvenile chronic arthritis and attention deficit-hyperactivity disorder [24, 56]. This may surprise many people working outside otolaryngology who may view these common ear and throat conditions as trivial. It seems that parents rate the burden of ill health associated with these conditions as far from trivial. It is precisely because they allow us to put our clinical work in such a

context that otolaryngologists should find generic HRQOL measures useful in their research.

The otolaryngologist wishing to identify a generic HRQOL measure for use in a research setting could select any of the measures studied with some confidence, then, since all have been shown to perform reasonably well in the otolaryngology setting. Certainly, there does not seem to be any immediately pressing need for us to be designing new generic measures, as the MRC had to do for the TARGET trial. The generic measures now available are sufficiently informative that, used in combination with a disease-specific instrument, any one of them is likely to be more than adequate. For studies largely involving pre-school children, TAPQOL is the obvious choice. If older children are to be studied, the CHQ has the advantage of being the most widely used, although TACQOL should be considered for its potential to be more sensitive to otolaryngology issues. The HUI mark III seems to be a good choice for economic studies in otolaryngology, studies which will become increasingly important for setting priorities in a health service where funding is always going to be limited. It may also be useful where issues of hearing are important, since both the CHQ and TAPQOL have demonstrated a lack of sensitivity in this area.

There are some situations where the standard generic HRQOL instruments may not be ideal. New instruments have been evaluated in this thesis, one for use as a post-intervention measure of HRQOL benefit, the other to assess the impact

on the family. Both these instruments have been shown to perform well and will hopefully prove useful to otolaryngologists conducting research in these areas. Time and further experience are needed to refine them further, and studies of test-retest reliablity and interobserver variation (especially variation in scores between mother-and-father pairs of respondents) are clearly required. In both cases, a very high degree of internal consistency raises the possibility that it may be possible to produce a much shorter version of the instrument. This would be, however, at the cost of some richness in the data that may prove more useful for teasing out factors within it. Further work will demonstrate how informative these factors are.

Since the parental viewpoint is the one that informs most decisions about a child's health care, the impact of a disease on the child's family is very important. Hitherto, it has been difficult to make any assessment of such matters, but we have shown here that "family impact" is a coherent construct that can be measured in a practical and informative way. It is hoped that this measure can now be used in studies on the family impact of various conditions (in addition to the impact on the child) and the way this distinction informs and influences surgical decision making.

With the GCBI it has been possible to produce a measure which is centred around the priorities of parents (as well as health care workers) and which should be applicable to a wide range of conditions in paediatric medicine. It provides a practical way to measure benefit, with none of the drawbacks of a

before-and-after approach. It has been shown to be sensitive to improvements after surgery and free from any obvious effect of expectation bias (being appropraitely anchored at zero for interventions not thought to have been beneficial). It thus shows promise and will hopefully become at least as widely used as its adult counterpart.

Whatever the clinical situation under study, it should be possible for the researcher in paediatric otolaryngology to select a suitable outcome measure from the range available and thereby effectively evaluate the benefit of the work we do.

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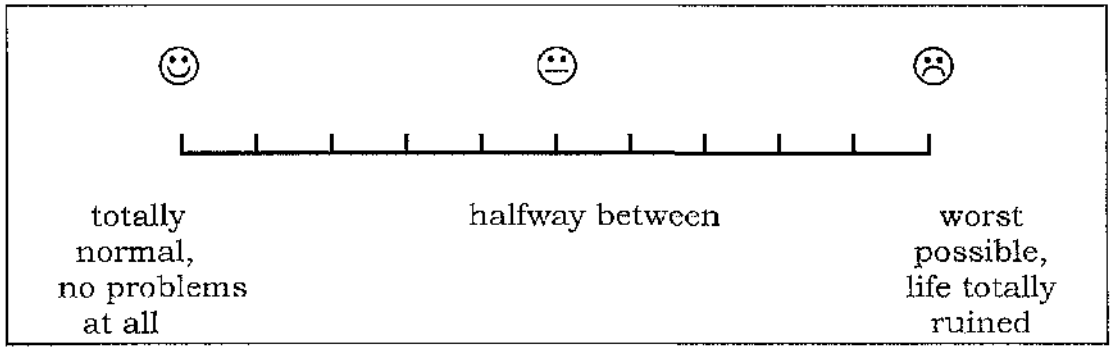
Appendix – the instruments used

1. The visual analogue scale
2. The Glasgow Children's Benefit Inventory
3. Quality of Family Life
4. The Health Utilities Index mark III
5. The Child Health Questionnaire Parent Form 50
6. TACQOL
7. TAPQOL
8. The clinical data questionnaires posted out with the GCBI, including RHD-4

Children's Quality of Life Study

Please put a mark somewhere on the line below to show how much your child's life has been affected overall by their ear, nose and throat problems over the last 3 months.



Glasgow Children's Benefit Inventory

In this questionnaire, we are interested to know how much change you think there has been in your child's general condition since his or her operation.

1. Has your child's operation made his/her overall life better or worse?

Much better	A little better	No change	A little worse	Much worse

2. Has your child's operation affected the things he/she does?

Much better	A little better	No change	A little worse	Much worse

3. Has your child's operation made his/her behaviour better or worse?

Much better	A little better	No change	A little worse	Much worse

4. Has your child's operation affected his/her progress and development?

Much better	A little better	No change	A little worse	Much worse

5. Has your child's operation affected how lively he/she is during the day?

Much better	A little better	No change	A little worse	Much worse

6. Has your child's operation affected how well he/she sleeps at night?

Much better	A little better	No change	A little worse	Much worse

7. Has your child's operation affected his/her enjoyment of food?

Much better	A little better	No change	A little worse	Much worse

8. Has your child's operation affected how self-conscious he/she is with other people?

Much better	A little better	No change	A little worse	Much worse

9. Has your child's operation affected how well he/she gets on with the rest of the family?

Much better	A little better	No change	A little worse	Much worse

10. Has your child's operation affected his/her ability to spend time and have fun with friends?

Much better	A little better	No change	A little worse	Much worse

11. Has your child's operation affected how embarrassed he/she is with other people?

Much better	A little better	No change	A little worse	Much worse

12. Has your child's operation affected how easily distracted he/she has been?

Much better	A little better	No change	A little worse	Much worse

13. Has your child's operation affected his/her learning?

Much better	A little better	No change	A little worse	Much worse

14. Has your child's operation affected the amount of time he/she has had to be off nursery, playgroup or school?

Much better	A little better	No change	A little worse	Much worse

15. Has your child's operation affected his/her ability to concentrate on a task?

Much better	A little better	No change	A little worse	Much worse

16. Has your child's operation affected how frustrated and irritable he/she is?

Much better	A little better	No change	A little worse	Much worse

17. Has your child's operation affected how he/she feels about himself/herself?

Much better	A little better	No change	A little worse	Much worse

18. Has your child's operation affected how happy and content he/she is?

Much better	A little better	No change	A little worse	Much worse

19. Has your child's operation affected his/her confidence?

Much better	A little better	No change	A little worse	Much worse

20. Has your child's operation affected his/her ability to care for himself/herself as well as you think they should, such as washing, dressing and using the toilet?

Much better	A little better	No change	A little worse	Much worse

21. Has your child's operation affected his/her ability to enjoy leisure activities such as swimming and sports, and general play?

Much better	A little better	No change	A little worse	Much worse

22. Has your child's operation affected how prone he/she is to catch colds or infections?

Much better	A little better	No change	A little worse	Much worse

23. Has your child's operation affected how often he/she needs to visit a doctor?

Much better	A little better	No change	A little worse	Much worse

24. Has your child's operation affected how much medication he/she has needed to take?

Much better	A little better	No change	A little worse	Much worse

Thank you for taking the time to complete this questionnaire!

QUALITY OF FAMILY LIFE

We would like to find out how your child's ear, nose and throat problems have affected your family. Please answer the questions for your family as a whole. By family, we mean the people who normally live with you, or look after your child, such as grandparents, brothers and sisters.

You may want to talk to other members of your family before answering.

Everything you say will be treated confidentially.

The questions in Section A are about who is in your family

Section B asks about how your family feels about things now

Thank you for taking the time to fill in this questionnaire.

SECTION A

Not all families are the same. Please could you tell us who is in your immediate family. Usually, these will be the people who live with you.

We don't want to know their names, just how they are related to you and how old they are.

The example shows a family of four, a woman, her husband, their son and the husband's mother.

EXAMPLE:

MYSELF	**Age ....50.....**
My .....Husband.....	**Age .....52.....**
My .....Son.....	**Age .....20.....**
My .....Mother-in-law.....	**Age .....74.....**

Now please fill in the box below for yourself and your family.

YOUR FAMILY:

The one who is filling in the questionnaire:

MYSELF **Age .....**

The child who is coming to the ear, nose and throat clinic:

My ..... **Age .....**

The rest of the family

My ..... **Age .....**

My ..... **Age .....**

My ..... **Age .....**

My ..... **Age .....**

My ..... **Age .....**

My ..... **Age .....**

SECTION B

Taking into account your child's ear, nose and throat problems over the last four weeks, please answer the following questions about how your family feels.

Please answer the questions for your family as a whole

Taking into account your child's ear, nose and throat problems

1-...how much enjoyment does your family get from going out together?

A great deal of enjoyment	Quite a lot of enjoyment	Some enjoyment	Not much enjoyment	No enjoyment or very little enjoyment
( )	( )	( )	( )	( )

2-...is your family restricted in going out together?

Not at all restricted	Slightly restricted	Moderately restricted	Severely restricted	Very severely restricted
( )	( )	( )	( )	( )

3- ...how much of an effort is it for your family to get ready in the morning?

No effort at all	Only a little effort	Some effort	Quite a lot of effort	A great deal of effort
( )	( )	( )	( )	( )

4-...how confident is your family that it has enough time to do all the household activities it has to do (e.g. chores, odd jobs)?

Not confident at all	Not very confident	Somewhat confident	Quite confident	Very confident
( )	( )	( )	( )	( )

5-...how satisfied is your family with the support it receives from people around it (e.g. from friends, family and others)?

Very satisfied	Quite satisfied	Somewhat satisfied	Not very satisfied	Not satisfied at all
( )	( )	( )	( )	( )

Taking into account your child's ear, nose and throat problems

6-...how confident is your family that it is coping with life in general?

Not confident at all	Not very confident	Somewhat confident	Quite confident	Very confident
( )	( )	( )	( )	( )

Taking into account your child's ear, nose and throat problems

7-...how confident is your family that it will be able to cope with life in general in the future?				
Not confident at all	Not very confident	Somewhat confident	Quite confident	Very confident
( )	( )	( )	( )	( )

8-...how much enjoyment does your family get from watching TV together				
A great deal of enjoyment	Quite a lot of enjoyment	Some enjoyment	Not much enjoyment	No enjoyment or very little enjoyment
( )	( )	( )	( )	( )

9-...how confident is your family that it has enough time to do all the social and leisure activities it would like to do (e.g. entertaining, visiting friends, hobbies, sport)?				
Not confident at all	Not very confident	Somewhat confident	Quite confident	Very confident
( )	( )	( )	( )	( )

10-...how much enjoyment does your family get from having meals together at home				
No enjoyment or very little enjoyment	Not much enjoyment	Some enjoyment	Quite a lot of enjoyment	A great deal of enjoyment
( )	( )	( )	( )	( )

11-...how easy or difficult is it for your family to come to an agreement?				
Very easy	Easy	Neither easy nor difficult	Difficult	Very difficult
( )	( )	( )	( )	( )

12-...how much do other people interfere in your family's life/				
Don't interfere at all	Interfere just a little	Interfere somewhat	Interfere quite a lot	Interfere a great deal
( )	( )	( )	( )	( )

13-...how much enjoyment does your family get from going away on holiday together?				
No enjoyment or very little enjoyment	Only a little enjoyment	Some enjoyment	Quite a lot of enjoyment	A great deal of enjoyment
( )	( )	( )	( )	( )

Taking into account your child's ear, nose and throat problems

14-...is your family restricted in its choice of holidays?				
Very severely restricted	Severely restricted	Moderately restricted	Slightly restricted	Not at all restricted
( )	( )	( )	( )	( )

15-...how much does your family feel under pressure?				
Under no pressure	Under very little pressure	Under some pressure	Under quite a lot of pressure	Under a great deal of pressure
( )	( )	( )	( )	( )

16-...how worried is your family about the well-being of your child when you are not together?				
Not worried at all	Just a little worried	Somewhat worried	Worried quite a lot	Very worried
( )	( )	( )	( )	( )

17-...is your family satisfied with its achievements (e.g. in work, school, sports or hobbies)?				
Not satisfied at all	Not very satisfied	Somewhat satisfied	Quite satisfied	Very satisfied
( )	( )	( )	( )	( )

18-..is any stress caused when including your child in family activities?				
No stress at all	Only a little stress	Some stress	Quite a lot of stress	A great deal of stress
( )	( )	( )	( )	( )

19-...which of the following statements best describes your family's view of the future?				
Not confident at all	Not very confident	Somewhat confident	Quite confident	Very confident
( )	( )	( )	( )	( )

20-...is your family confident that it has enough money to keep up its standard of living?				
Not confident at all	Not very confident	Somewhat confident	Quite confident	Very confident
( )	( )	( )	( )	( )

Taking into account your child's ear, nose and throat problems

21-...how satisfied is your family that its needs are being met?				
Not satisfied at all	Not very satisfied	Somewhat satisfied	Quite satisfied	Very satisfied
( )	( )	( )	( )	( )

22-...is any embarrassment caused when including your child in family activities?				
No embarrassment at all	Only a little embarrassment	Some embarrassment	Quite a lot of embarrassment	A great deal of embarrassment
( )	( )	( )	( )	( )

23-...how much does your family understand about your child's ear, nose and throat problems?				
As much as we would like	Not quite as much as we would like	Less than we would like	Much less than we would like	Very much less than we would like
( )	( )	( )	( )	( )

24-...how much control does your family have over the way it lives its life?				
No control or very little control	Not much control	Some control	Quite a lot of of control	Complete or nearly complete control
( )	( )	( )	( )	( )

25-...how much enjoyment does your family get from spending time together at home (e.g. talking, playing games)?				
No enjoyment or very little enjoyment	Not much enjoyment	Some enjoyment	Quite a lot of enjoyment	A great deal of enjoyment
( )	( )	( )	( )	( )

26- how happy is your family?				
Very happy	Happy	Somewhat happy	Not very happy	Not happy at all
( )	( )	( )	( )	( )

We have dealt with some of the aspects of family life and activities which may be affected by a child with ear, nose and throat problems. We would now like you to think of any other aspects or activities, which are important to <u>your</u> family, which might be affected. Please write them in the shaded boxes and then tick the answer which best describes how much your family is affected.

27.				
Cannot manage to do at all ( )	**Affected very much** ( )	**Affected quite a lot** ( )	**Affected somewhat** ( )	**Affected only slightly** ( )

28.				
Cannot manage to do at all ( )	**Affected very much** ( )	**Affected quite a lot** ( )	**Affected somewhat** ( )	**Affected only slightly** ( )

Thank you very much for your help

HEALTH UTILITIES INDEX MARK 2 AND MARK 3 (HUI2/3)
15-ITEM QUESTIONNAIRE FOR
PARENT-COMPLETED
4 WEEK HEALTH STATUS ASSESSMENT

Instructions: This questionnaire contains a set of questions which ask about various aspects of your child's health. When answering these questions please think about your child's health and ability to do things on a day-to-day basis, during the past 4 weeks. To define the 4 week period, please think about what the date was 4 weeks ago and recall the major events that you have experienced during this period. Please focus your answers on your child's overall abilities, disabilities and how he or she felt during the past 4 weeks.

You may feel that some of these questions do not apply to your child, but it is important that we ask the same questions of everyone. Also, a few questions are similar; please excuse the apparent overlap and answer each question independently.

Please read each question and consider your answers carefully. For each question, please select one answer that best describes your child's level of ability or disability during the past 4 weeks. Please indicate the selected answer by circling the letter (a,b,c,.....) beside the answer.

All information you provide is confidential. There are no right or wrong answers; what we want is your opinion about your child's abilities and feelings.

1. Which one of the following best describes your child's ability, during the past 4 weeks, to see well enough to read ordinary newsprint?

   a. Able to see well enough without glasses or contact lenses.

   b. Able to see well enough with glasses or contact lenses.

   c. Unable to see well enough even with glasses or contact lenses.

   d. Unable to see at all.

2. Which one of the following best describes your child's ability, during the past 4 weeks, to see well enough to recognise a friend on the other side of the street?

   a. Able to see well enough without glasses or contact lenses.

   b. Able to see well enough with glasses or contact lenses.

   c. Unable to see well enough even with glasses or contact lenses.

   d. Unable to see at all.

3. Which one of the following best describes your child's ability, during the past 4 weeks, to hear what was said in a group conversation with at least three other people?

   a. Able to hear what was said without a hearing aid.

   b. Able to hear what was said with a hearing aid.

   c. Unable to hear what was said even with a hearing aid.

   d. Unable to hear what was said, but did not wear a hearing aid.

   e. Unable to hear at all.

4. Which one of the following best describes your child's ability, during the past 4 weeks, to hear what was said in a conversation with one other person in a quiet room?

   a. Able to hear what was said without a hearing aid.

   b. Able to hear what was said with a hearing aid.

   c. Unable to hear what was said even with a hearing aid.

   d. Unable to hear what was said, but did not wear a hearing aid.

   e. Unable to hear at all.

5. Which one of the following best describes your child's ability, during the past 4 weeks, to be understood when speaking his or her own language with people who do not know him or her?

   a. Able to be understood completely.

   b. Able to be understood partially.

   c. Unable to be understood.

   d. Unable to speak at all.

6. Which one of the following best describes your child's ability, during the past 4 weeks, to be understood when speaking with people who know him or her well?

   a. Able to be understood completely.

   b. Able to be understood partially.

   c. Unable to be understood.

   d. Unable to speak at all.

7. Which one of the following best describes how your child has been feeling during the past 4 weeks?

   a. Happy and interested in life.

   b. Somewhat happy.

   c. Somewhat unhappy.

   d. Very unhappy.

   e. So unhappy that life was not worthwhile.

8. Which one of the following best describes the pain and discomfort your child has experienced during the past 4 weeks?

   a. Free of pain and discomfort.

   b. Mild to moderate pain or discomfort that prevented no activities.

   c. Moderate pain or discomfort that prevented a few activities.

   d. Moderate to severe pain or discomfort that prevented some activities.

   e. Severe pain or discomfort that prevented most activities.

9. Which one of the following best describes your child's ability, during the past 4 weeks, to walk? Note: Walking equipment refers to mechanical supports such as braces, a cane, crutches or a walker.

a. Able to walk around the neighbourhood without difficulty, and without walking equipment.

b. Able to walk around the neighbourhood with difficulty; but did not require walking equipment or the help of another person.

c. Able to walk around the neighbourhood with walking equipment, but without the help of another person.

d. Able to walk only short distances with walking equipment, and required a wheelchair to get around the neighbourhood.

e. Unable to walk along, even with walking equipment. Able to walk short distances with the help of another person, and required a wheelchair to get around the neighbourhood.

f. Unable to walk at all.

10. Which one of the following best describes your child's ability, during the past 4 weeks, to use is or her hands and fingers?
Note: Special tools refer to hooks for buttoning clothes, gripping devices for opening jars or lifting small items, and other devices to compensate for limitations of hands or fingers.

a. Full use of two hands and ten fingers.

b. Limitations in the use of hands or fingers, but did not require special tools or the help of another person.

c. Limitations in the use of hands or fingers, independent with the use of special tools (did not require the help of another person).

d. Limitations in the use of hands or fingers, required the help of another person for some tasks (not independent even with use of special tools).

e. Limitations in the use of hands or fingers, required the help of another person for most tasks (not independent even with use of special tools).

f. Limitations in the use of hands or fingers, required the help of another person for all tasks (not independent even with use of special tools).

11. Which one of the following best describes your child's ability, during the past 4 weeks, to remember things?

a. Able to remember most things.

b. Somewhat forgetful.

c. Very forgetful.

d. Unable to remember anything at all.

12. Which one of the following best describes your child's ability, during the past 4 weeks, to think and solve day to day problems?

a. Able to think clearly and solve day to day problems.

b. Had a little difficulty when trying to think and solve day to day problems.

c. Had some difficulty when trying to think and solve day to day problems.

d. Had great difficulty when trying to think and solve day to day problems.

e. Unable to think or solve day to day problems.

13. Which one of the following best describes your child's ability, during the past 4 weeks, to perform basic activities.

a. Eat, bathe, dress and use the toilet normally.

b. Eat, bathe, dress and use the toilet independently with difficulty.

c. Required mechanical equipment to eat, bathe, dress or use the toilet independently.

d. Required the help of another person to eat, bathe, dress or use the toilet.

14. Which one of the following best describes how your child has been feeling during the past 4 weeks?

a. Generally happy and free from worry.

b. Occasionally fretful, angry, irritable, anxious or depressed.

c. Often fretful, angry, irritable, anxious or depressed.

d. Almost always fretful, angry, irritable, anxious or depressed.

e. Extremely fretful, angry, irritable, anxious or depressed; to the point of needing professional help.

15. Which one of the following best describes the pain or discomfort your child has experienced during the past 4 weeks?

a. Free of pain and discomfort.

b. Occasional pain or discomfort. Discomfort relieved by non-prescription drugs or self-control activity without disruption of normal activities.

c. Frequency pain or discomfort. Discomfort relieved by oral medicines with occasional disruption of normal activities.

d. Frequency pain or discomfort; frequent disruption of normal activities. Discomfort required prescription narcotics for relief.

e. Severe pain or discomfort. Pain not relieved by drugs and constantly disrupted normal activities.

16. Overall, how would you rate your child's health during the past 4 weeks?

a. Excellent.

b. Very good.

c. Good

d. Fair.

e. Poor.

Child Health Questionnaire – Parent Report
CHQ-PF50

- INSTRUCTIONS -

1. This booklet asks about your child's health and well-being. Your individual answers will not be shared with anyone.

2. If you choose not to participate it will not affect the care you receive.

3. Answer the questions by marking the appropriate box ☑

4. Certain questions may look alike but each one is different. Some questions ask about problems your child may not have, but it's important for us to know that too. Please answer each question.

5. There are no right or wrong answers. If you are unsure how to answer a question, please give the best answer you can and make a comment in the margin.

6. All comments will be read, so please feel free to make as many as you wish.

SECTION 1: YOUR CHILD'S GENERAL HEALTH

1.1 In general, would you say <u>your child's health</u> is:

☐	☐	☐	☐	☐
Excellent	Very good	Good	Fair	Poor

SECTION 2: YOUR CHILD'S PHYSICAL ACTIVITIES

The following questions ask about physical activities your child might do during a day.

2.1 During the <u>past 4 weeks</u>, has your child been limited in any of the following activities due to <u>health problems</u>?

		Yes, limited a lot	Yes, limited somewhat	Yes, limited a little	No, not limited
a.	Doing things that take **a lot** of energy, such as playing football or running?	☐	☐	☐	☐
b.	Doing things that take **some** energy such as riding a bike or roller skating?	☐	☐	☐	☐
c.	Ability (physically) to get around the neighbourhood, playground or school?	☐	☐	☐	☐
d.	Walking 100 metres or climbing one flight of stairs?	☐	☐	☐	☐
e.	Bending, lifting, or stooping?	☐	☐	☐	☐
f.	Taking care of him/herself, that is, eating, dressing, bathing, or going to the toilet?	☐	☐	☐	☐

SECTION 3: YOUR CHILD'S EVERYDAY ACTIVITIES

3.1 During the <u>past 4 weeks</u>, has your child's schoolwork or activities with friends been limited in any of the following ways due to EMOTIONAL difficulties or problems with his/her BEHAVIOUR?

		Yes, limited a lot	Yes, limited somewhat	Yes, limited a little	No, not limited
a.	limited in the KIND of schoolwork or activities with friends he/she could do	☐	☐	☐	☐
b.	limited in the AMOUNT of time he/she could spend on schoolwork or activities with friends	☐	☐	☐	☐
c.	limited in PERFORMING schoolwork or activities with friends (it took extra effort)	☐	☐	☐	☐

3.2 During the <u>past 4 weeks</u>, has your child's schoolwork or activities with friends been limited in any of the following ways due to problems with his/her PHYSICAL health?

		Yes, limited a lot	Yes, limited somewhat	Yes, limited a little	No, not limited
a.	limited in the KIND of schoolwork or activities with friends he/she could do	☐	☐	☐	☐
b.	limited in the AMOUNT of time he/she could spend on schoolwork or activities with friends	☐	☐	☐	☐

SECTION 4: PAIN

4.1 During the past 4 weeks, how much bodily pain or discomfort has your child had?

☐	☐	☐	☐	☐	☐
None	Very mild	Mild	Moderate	Severe	Very severe

4.2 During the past 4 weeks, how often has your child had bodily pain or discomfort?

☐	☐	☐	☐	☐	☐
None of the time	Once or twice	A few times	Fairly often	Very often	Every/almost every day

SECTION 5: BEHAVIOUR

Below is a list of items that describe children's behaviour or problems they sometimes have.

5.1 How often during the past 4 weeks did each of the following statements describe your child?

		Very often	Fairly often	Sometimes	Almost never	Never
a.	argued a lot	☐	☐	☐	☐	☐
b.	had difficulty concentrating or paying attention	☐	☐	☐	☐	☐
c.	not told the truth	☐	☐	☐	☐	☐
d.	taken things which didn't belong to them	☐	☐	☐	☐	☐
e.	had tantrums or a hot temper	☐	☐	☐	☐	☐

5.2 Compared to other children your child's age, in general would you say his/her behaviour is:

☐	☐	☐	☐	☐
Excellent	Very good	Good	Fair	Poor

SECTION 6: WELL-BEING

The following phrases are about children's moods

6.1 During the past 4 weeks, how much of the time do you think your child:

		All of the time	Most of the time	Some of the time	A little of the time	None of the time
a.	felt like crying?	☐	☐	☐	☐	☐
b.	felt lonely?	☐	☐	☐	☐	☐
c.	acted nervous?	☐	☐	☐	☐	☐
d.	acted bothered or upset?	☐	☐	☐	☐	☐
e.	acted cheerful?	☐	☐	☐	☐	☐

SECTION 7: SELF-ESTEEM

The following ask about your child's satisfaction with self, school, and others. It may be helpful if you keep in mind how other children your child's age might feel about these areas.

7.1 During the past 4 weeks, how satisfied do you think your child has felt about:

		Very satisfied	Somewhat satisfied	Neither satisfied nor dissatisfied	Somewhat dissatisfied	Very dissatisfied
a.	his/her school ability?	☐	☐	☐	☐	☐
b.	his/her athletic ability?	☐	☐	☐	☐	☐
c.	his/her friendships?	☐	☐	☐	☐	☐
d.	his/her looks/appearance?	☐	☐	☐	☐	☐
e.	his/her family relationships?	☐	☐	☐	☐	☐
f.	his/her life overall?	☐	☐	☐	☐	☐

SECTION 8: YOUR CHILD'S HEALTH

The following statements are about health in general

8.1 How true or false is each of these statements for your child?

		Definitely true	Mostly true	Don't know	Mostly false	Definitely false
a.	My child seems to be less healthy than other children I know.	☐	☐	☐	☐	☐
b.	My child has never been seriously ill.	☐	☐	☐	☐	☐
c.	When there is something going around my child usually catches it.	☐	☐	☐	☐	☐
d.	I expect my child will have a very healthy life.	☐	☐	☐	☐	☐
e.	I worry more about my child's health than other people worry about their children's health.	☐	☐	☐	☐	☐

8.2 Compared to one year ago, how would you rate your child's health now:

☐	☐	☐	☐	☐
Much better now than 1 year ago	Somewhat better now than 1 year ago	About the same now as 1 year ago	Somewhat worse now than 1 year ago	Much worse now than 1 year ago

SECTION 9: YOU AND YOUR FAMILY

9.1 During the <u>past 4 weeks</u>, how MUCH emotional worry or concern did each of the following cause YOU?

		None at all	A little bit	Somewhat	A lot	A great deal
a.	Your child's physical health	☐	☐	☐	☐	☐
b.	Your child's emotional well-being or behaviour	☐	☐	☐	☐	☐
c.	Your child's attention or learning abilities	☐	☐	☐	☐	☐

9.2 During the <u>past 4 weeks</u>, were you LIMITED in the amount of time YOU had for your own needs because of:

		Yes, limited a lot	Yes, limited somewhat	Yes, limited a little	No, not limited
a.	Your child's physical health?	☐	☐	☐	☐
b.	Your child's emotional well-being or behaviour?	☐	☐	☐	☐
c.	Your child's attention or learning abilities?	☐	☐	☐	☐

9.3 During the past 4 weeks, <u>how often</u> has your child's <u>health or behaviour</u>:

		Very often	Fairly often	Sometimes	Almost never	Never
a.	limited the types of activities you could do as a family?	☐	☐	☐	☐	☐
b.	interrupted various everyday family activities (eating meals, watching tv)?	☐	☐	☐	☐	☐
c.	limited your ability as a family to "get-up and go" on a moment's notice?	☐	☐	☐	☐	☐
d.	caused tension or conflict in your home?	☐	☐	☐	☐	☐
e.	been a source of disagreements or arguments in your family?	☐	☐	☐	☐	☐
f.	caused you to cancel or change plans (personal or work) at the last minute?	☐	☐	☐	☐	☐

9.4 Sometimes families may have difficulty getting along with one another. They do not always agree and they may get angry. In general, how would you rate your family's ability to get along with one another?

☐	☐	☐	☐	☐
Excellent	Very good	Good	Fair	Poor

THANK YOU FOR YOUR PARTICIPATION!

TACQOL

Questionnaire

for parents/carers of children aged 6 to 15

Would you please answer the following questions first?

Is the child in question a boy or a girl?	θ boy	θ girl	
What is the child's date of birth?	_____ (day)	_____ (month)	_____ (year)
On what date was this questionnaire completed?	_____ (day)	_____ (month)	_____ (year)

Number: [_ _ _ _ _ _ _ _ _ _ _ _]

Dear parents

We wish to know how your child has been in recent weeks.

On the pages which follow, you will find a number of questions.
There are a number of answers for each question.
Choose the answer which is the most appropriate for your child and place a cross in the box alongside that answer.

For example (you do not need to answer this question):

Has your child had headaches?.

θ never θ occasionally θ often

1

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Has your child had earaches or sore throats?.

θ never θ occasionally θ often

2

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

If your child has **not** suffered from headaches **at all** in recent weeks, place a cross in the box next to 'never'. You can then go on to the next question about sore throats as in the example above.

If your child had a headache "occasionally" or "often", place a cross in the appropriate box. Below these boxes, you find the words: '**At that time, my child felt:**' You then cross the box stating how your child felt when he or she had a headache.

For example:

Has your child had headaches?.

θ never θ occasionally θ often

1

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

You then proceed to the next question

Pain and symptoms in recent weeks

Try to remember how your child was in recent weeks

Has your child had earaches or sore throats? θ never θ occasionally θ often

1

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Has your child had stomach-aches or abdominal pain? θ never θ occasionally θ often

2

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Has your child had headaches? θ never θ occasionally θ often

3

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Has your child been dizzy? θ never θ occasionally θ often

4

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Has your child felt sick/nauseous? θ never θ occasionally θ often

5

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Was your child tired? θ never θ occasionally θ often

6

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Was your child sleepy? θ never θ occasionally θ often

7

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Was your child dozy/lethargic? θ never θ occasionally θ often

8

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child suffer from pain or other symptoms? θ never θ occasionally θ often

9

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

What sort of pains or symptoms?

Only if your child suffered from pains or other symptoms in recent weeks:
What do you think caused those pains or those symptoms?

10

Things which your child had difficulty with in recent weeks

Try to remember how your child was in recent weeks. Did he or she have …

Difficulty with running? θ never θ often θ often

11

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with walking? θ never θ occasionally θ often

12

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with standing? θ never θ occasionally θ often

13

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty walking downstairs? θ never θ occasionally θ often

14

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with playing? θ never θ occasionally θ often

15

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with running or walking for long periods, with stamina? θ never θ occasionally θ often

16

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with balance? θ never θ occasionally θ often

17

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with doing things handily or quickly? θ never θ occasionally θ often

18

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Only if your child had problems of this kind in recent weeks:

What do you think caused these problems?

19

Things which your child had difficulty with in recent weeks

Try to remember how your child was in recent weeks. Did he or she have …

Difficulty with going to school on his/her own? θ never θ occasionally θ often

20

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty washing himself/herself? θ never θ occasionally θ often

21

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty getting dressed on his/her own? θ never θ occasionally θ often

22

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty going to the lavatory on his/her own? θ never θ occasionally θ often

23

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with eating or drinking on his/her own? θ never θ occasionally θ often

24

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with sports or going out to play on his/her own? θ never θ occasionally θ often

25

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with doing hobbies on his/her own? θ never θ occasionally θ often

26

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with riding a bicycle? θ never θ occasionally θ often

27

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Only if your child had problems of this kind in recent weeks:

What do you think caused these problems?

28

Things which your child had difficulty with in recent weeks

Try to remember how your child was in recent weeks. Did he or she have ...

Difficulty with paying attention, concentrating? θ never θ occasionally θ often

29

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty understanding schoolwork? θ never θ occasionally θ often

30

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty understanding what others said? θ never θ occasionally θ often

31

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with arithmetic? θ never θ occasionally θ often

32

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with reading? θ never θ occasionally θ often

33

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with writing? θ never θ occasionally θ often

34

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with learning? θ never θ occasionally θ often

35

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty in saying what he/she meant? θ never θ occasionally θ often

36

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Only if your child had problems of this kind in recent weeks:

What do you think caused these problems?

37

Dealings with other children and with you in recent weeks

Try to remember how your child was in recent weeks.

My child was able to play or talk happily with other children. θ yes θ too little θ never

38

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

My child was able to stand up for himself/herself with other children. θ yes θ too little θ never

39

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Other children asked my child to play with them. θ yes θ too little θ never

40

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

My child was at ease with other children. θ yes θ too little θ never

41

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

My child was able to play or talk happily with us – the parent(s). θ yes θ too little θ never

42

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

My child was incommunicative or quiet with us – the parent(s). θ never θ occasionally θ often

43

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

My child was restless or impatient with us – the parent(s). θ never θ occasionally θ often

44

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

My child was defiant with us – the parent(s). θ never θ occasionally θ often

45

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

If things were not always satisfactory in dealings with other children or with you:
What do you think was the reason?

46

In recent weeks my child felt ...

Joyful 47	0 never	0 occasionally	0 often	**Relaxed** 55	0 never	0 occasionally	0 often
Sad 48	0 never	0 occasionally	0 often	**Aggressive** 56	0 never	0 occasionally	0 often
In good spirits 49	0 never	0 occasionally	0 often	**Happy** 57	0 never	0 occasionally	0 often
Angry 50	0 never	0 occasionally	0 often	**Short-tempered** 58	0 never	0 occasionally	0 often
Contented 51	0 never	0 occasionally	0 often	**Confident** 59	0 never	0 occasionally	0 often
Worried 52	0 never	0 occasionally	0 often	**Jealous** 60	0 never	0 occasionally	0 often
Enthusiastic 53	0 never	0 occasionally	0 often	**Cheerful** 61	0 never	0 occasionally	0 often
Gloomy 54	0 never	0 occasionally	0 often	**Anxious** 62	0 never	0 occasionally	0 often

If your child did not always feel fine in recent weeks:
What was the reason?

63

This is the end of the questionnaire
Thank you for completing it!

TAPQOL

Questionnaire

for parents of children aged 1 to 5

Would you please answer the following questions first?

Is the child in question a boy or a girl?	θ boy	θ girl	
What is the child's date of birth?	_____ (day)	_____ (month)	_____ (year)
On what date was this questionnaire completed?	_____ (day)	_____ (month)	_____ (year)

Number: [_ _ _ _ _ _ _ _ _ _ _ _ _]

INSTRUCTIONS

Dear Sir/Madam,

The questions in this questionnaire relate to all kinds of different aspects of your child's health.
You can answer the questions by ticking the answer which best describes your child.

For example:

In the last three months, has your child had ..

Ear-ache ☒ never θ occasionally θ often

1

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

If things were not entirely satisfactory, you are also asked how your child felt when there was a problem.
So, if you say that your child had ear-ache 'occasionally' or 'often', you can state, in the second part of the question, how your child felt at that time.

For example:

In the last three months, has your child had ..

Ear-ache θ never ☒ occasionally θ often

1

At that time, my child felt:

θ fine θ not so good ☒ quite bad θ bad

In the last three months, has your child had ..

Stomach-ache or abdominal pain θ never θ occasionally 0 often

1

At that time, my child felt:

0 fine θ not so good θ quite bad θ bad

Colic θ never θ occasionally θ often

2

At that time, my child felt:

0 fine θ not so good θ quite bad θ bad

Eczema θ never θ occasionally θ often

3

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Itchiness 0 never θ occasionally θ often

4

At that time, my child felt:

θ fine θ not so good 0 quite bad θ bad

Dry skin θ never θ occasionally θ often

5

At that time, my child felt:

θ fine θ not so good θ quite bad 0 bad

Bronchitis θ never θ occasionally θ often

6

At that time, my child felt:

θ fine 0 not so good θ quite bad θ bad

Difficulty with breathing or lung problems θ never θ occasionally 0 often

7

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

In the last three months, has your child been ..

Short of breath

θ never θ occasionally θ often

8

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Nauseous

θ never θ occasionally θ often

9

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

How did your child sleep in the last three months?

Did your child sleep restlessly?

θ never θ occasionally θ often

10

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Was your child awake at night?

θ never θ occasionally θ often

11

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child cry at night?

θ never θ occasionally θ often

12

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child have difficulty sleeping through the night?

θ never θ occasionally θ often

13

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

How did your child eat and drink in the last three months?

Was your child's appetite poor? θ never θ occasionally θ often

14

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child vomit after eating? θ never θ occasionally θ often

15

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child have difficulty swallowing food? θ never θ occasionally θ often

16

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child have difficulty eating enough? θ never θ occasionally θ often

17

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child refuse to eat? θ never θ occasionally θ often

18

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Did your child refuse to drink? θ never θ occasionally θ often

19

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Your child's behaviour in the last three months

My child was short-tempered 20	θ never	θ occasionally	θ often
My child was aggressive 21	θ never	θ occasionally	θ often
My child was irritable 22	θ never	θ occasionally	θ often
My child was angry 23	θ never	θ occasionally	θ often
My child was restless or impatient with me 24	θ never	θ occasionally	θ often
My child was defiant/awkward with me 25	θ never	θ occasionally	θ often
I could not manage my child 26	θ never	θ occasionally	θ often

How was your child in the last three months

In good spirits 27	θ never	θ occasionally	θ often
Cheerful 28	θ never	θ occasionally	θ often
Happy 29	θ never	θ occasionally	θ often
Frightened 30	θ never	θ occasionally	θ often
Tense 31	θ never	θ occasionally	θ often
Anxious 32	θ never	θ occasionally	θ often
Energetic 33	θ never	θ occasionally	θ often
Active 34	θ never	θ occasionally	θ often
Lively 35	θ never	θ occasionally	θ often

If your child is aged below eighteen months, you do not have to complete the rest of this questionnaire.

If your child is older than eighteen months, you should continue with the questions on the following pages.

How was your child's behaviour with other children in the last three months?

My child was able to play happily with other children θ never θ occasionally θ often

36

My child was at ease with other children θ never θ occasionally θ often

37

My child was confident with other children θ never θ occasionally θ often

38

In the last three months, did your child have, compared to other children of the same age ...

Difficulty with walking? θ no θ yes, a little θ yes, a lot θ cannot walk

39

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with running? θ no θ yes, a little θ yes, a lot θ cannot walk

40

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with walking up stairs without help? θ no θ yes, a little θ yes, a lot θ cannot walk

41

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty with balance? θ no θ yes, a little θ yes, a lot θ cannot walk

42

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

In the last three months, did your child have, compared to other children of the same age ...

Difficulty in understanding what others said? θ never θ occasionally θ often

43

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty in talking clearly? θ never θ occasionally θ often

44

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty in saying what he/she meant? θ never θ occasionally θ often

45

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

Difficulty in making it clear what he/she wanted? θ never θ occasionally θ often

46

At that time, my child felt:

θ fine θ not so good θ quite bad θ bad

This is the end of the questionnaire.

Thank you for completing it!

Please tell us how things have been since your child's operation, by ticking one box for each question.

1. Since my child's grommet operation, he or she...

- ❑ has had no ear infections at all
- ❑ has had some ear infections, but not as many as before the operation
- ❑ has had as many ear infections as they had before the operation

- ❑ never really had ear infections before the operation anyway

2. Since my child's grommet operation, he or she...

- ❑ has had much better hearing and speech
- ❑ has had no improvement in hearing and speech

- ❑ never really had a problem with hearing or speech before the operation anyway

3. Did your child suffer any complications or problems after the operation?

- ❑ No
- ❑ Yes Please describe_____ _____

4. Overall, how satisfied are you with the decision to put grommets in your child's ears?

- ❑ Very happy
- ❑ Happy
- ❑ Not sure either way
- ❑ Unhappy
- ❑ Very unhappy

Reported Hearing Difficulty Scale (RHD-4)

For each of the four questions below, please tick one answer that best describes your child over the last few weeks

1) How would you describe your child's hearing?	❑ Normal ❑ Slightly below normal ❑ Poor ❑ Very poor ❑ Not sure
2) Has he/she misheard words when not looking at you?	❑ No ❑ Rarely ❑ Often ❑ Always ❑ Not sure
3) Has he/she had difficulty hearing when with a group of people?	❑ No ❑ Rarely ❑ Often ❑ Always ❑ Not sure
4) Has he/she asked for things to be repeated?	❑ No ❑ Rarely ❑ Often ❑ Always ❑ Not sure

Please tell us how things have been since your child's operation, by ticking one box for each question.

1. Since my child recovered from the operation to remove the tonsils, he or she...

- ❑ has had no sore throats at all
- ❑ has had some sore throats, but not as many as before the operation
- ❑ has had as many sore throats as they had before the operation
- ❑ never really had sore throats before the operation anyway

2. Did your child suffer any complications or problems after the operation?

- ❑ No
- ❑ Yes Please describe_____

3. Overall, how satisfied are you with the decision to remove your child's tonsils?

- ❑ Very happy
- ❑ Happy
- ❑ Not sure either way
- ❑ Unhappy
- ❑ Very unhappy

Appendix 2

The flow of patients through the studies described in Chapters 5 and 6.